Development of a condition-specific outcome measure for rare musculoskeletal conditions: A multisite study using stakeholder engagement

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Abbreviations

Abbreviation	Meaning
ACMC	Assessment of Capacity for Myoelectric Control
AHA	Assisting Hand Assessment
AMC	Arthrogryposis multiplex congenita
BOT-2	Bruininks-Oseretsky Second Edition
CNS	Central Nervous System
COPM	Canadian Occupational Performance Measure
CTT	Classical Test Theory
DASH	Disabilities of the Arm, Shoulder and Hand
ES	Effect Size
FDT	Functional Dexterity Test
GAS	Goal Attainment Scale
GMPM	Gross Motor Performance Measure
GPS	Graduate and Postdoctoral Studies
HRQOL	Health Related Quality of Life
ICC	Intra-class coefficient
ICF	International Classification of Functioning, Disability, and Health
ICF-CY	International Classification of Functioning, Disability, and Health – Child and Youth
iKT	Integrated Knowledge Translation
IRT	Item Response Theory
JTHFT	Jebsen-Taylor Hand Function Test
KT	Knowledge Translation
KTA	Knowledge-to-Action
MAUUL	Melbourne Assessment of Unilateral Upper Limb
MSK	Musculoskeletal
NGT	Nominal Group Technique
OI	Osteogenesis Imperfecta
OT	Occupational Therapist
PALS	Participation and Activity Limitation Survey
PDMS-2	Peabody Developmental Motor Scales Second Edition
PEDI	Pediatric Evaluation of Disability Inventory
PPEET	Public and Patient Engagement Evaluation Tool
PROMIS-UE	Patient Reported Outcome Measure Information System – Upper Extremity
PT	Physiotherapist
QUEST	Quality of Upper Extremity Skills Test
RMT	Rasch Measurement Theory
RUMM2030	Rasch Unidimensional Measurement Model Software 2030
SHAPE-UP	Shriners Hospitals Arthrogryposis Pediatric Evaluation – Upper Extremity
SHUEE	Shriners Hospital Upper Extremity Evaluation
SPOR	Strategy for Patient Outcome Research
UE	Upper Extremity
WHO	World Health Organization

Abstract

The upper extremity (UE), includes the shoulder, elbow, wrist, and hands, and plays a fundamental function for basic and instrumental daily life activities such as self-care, work, leisure, social communication, and play. Arthrogryposis Multiplex Congenita (AMC) is a term used to describe a group of congenital conditions characterized by joint contractures in two or more body areas. The literature surrounding AMC is sparse, particularly that of UE function in the field of rehabilitation. There currently exists no outcome measure specific for children and youth with AMC. Currently, clinicians working with children and youth with AMC utilize generic performance-based or patient-reported measures aimed at overall functional performance. However, generic measures, 1) were not developed for children with an UE deformity, 2) do not inform clinicians, youth and their families as to which specific joint or muscle is limiting functional performance, and 3) may not reflect the adaptations or compensatory strategies used by children with AMC when engaging in activities. When developing a measure specific to a population with a rare musculoskeletal (MSK) condition, clinicians', patients' and caregivers' perspectives and involvement is a crucial and necessary step. Additionally, researchers in the field of measurement in rehabilitation have acknowledged that in order to develop evidence-based measures, standardized metrics must be used. By using Rasch analysis, UE function can be measured by arranging items according to a difficulty continuum, while patients are scored according to an ability continuum.

The overall aim of this thesis was to develop an UE outcome measure specifically designed for children and youth with AMC using stakeholder engagement and Rasch analysis. To operationalize the overall aim of the doctoral thesis, four research studies were conducted and comprise three manuscripts in this thesis.

The first study was a scoping review (Manuscript 1) of the literature aimed at identifying which performance-based outcome measures are most frequently used to evaluate the UE function in pediatric rehabilitation. The second aim was to determine the link between constructs of the International Classification of Function, Disability, and Health (ICF) and meaningful concepts extracted from each measure. The results indicated that the participation and personal factors domains were misrepresented and lacking. Findings from this review reinforced the need for stakeholder engagement in the next phase of the thesis.

The second and third studies (Manuscript 2) aimed at determining the most clinically useful items to be included in an outcome measure for children with AMC. This was achieved through a nominal group technique with an adult with AMC and caregivers of children and youth with AMC as well as a three-round clinician survey. Based on stakeholder engagement, the preliminary version of the Shriners Hospitals Arthrogryposis Pediatric Evaluation – UPper extremity (SHAPE-UP) was developed.

The final study (Manuscript 3) consisted of administering the SHAPE-UP to 92 participants across six sites. In order to validate its use in AMC, the SHAPE-UP needed to be administered, and additional testing was needed to determine if the data fit the Rasch model. The aims of this study were 1) to evaluate the psychometric properties of the SHAPE-UP Task Completion scale using Rasch analysis and 2) determine the strength of the correlation between the number of joints involved and the Task Completion score. The SHAPE-UP now consists of a 7 tasks performance-based outcome measure where the Task Completion was modified to adopt a 3-point scoring. Initial psychometric evaluation using Rasch analysis provided evidence of content and construct validity as well as reliability.

The original contribution of this doctoral research is the development of the first AMC-specific pediatric upper extremity outcome measure, the SHAPE-UP informed by youth, families of children with AMC and clinician expert opinion.

Abrégé

Le membre supérieur (MS) comprend l'épaule, le coude, le poignet et les mains et joue un rôle fondamental dans les activités de base de la vie quotidienne telles que les soins personnels, le travail, les loisirs, la communication sociale et les jeux. L'arthrogrypose multiplex congénitale (AMC) est un terme utilisé pour décrire un groupe de conditions congénitales caractérisées par des contractures articulaires dans deux ou plusieurs parties du corps. Il n'existe actuellement aucune évaluation spécifique pour les enfants et les adolescents atteints d'AMC. Les cliniciens utilisent des mesures génériques basées sur les performances ou rapportées par les patients, qui visent à évaluer les performances fonctionnelles globales. Cependant, les mesures génériques 1) n'ont pas été développées pour les enfants présentant une déformation du MS, 2) n'informent pas les cliniciens, les jeunes et leurs familles sur l'articulation ou le muscle spécifique qui limite la performance fonctionnelle, et 3) peuvent ne pas refléter les adaptations ou les stratégies de compensation utilisées par les enfants atteints d'AMC lorsqu'ils participent à des activités. Lors du développement d'une mesure spécifique à une population souffrant d'une affection musculo-squelettique rare, le point de vue et l'implication des cliniciens, des patients et des soignants constituent une étape cruciale et nécessaire. L'analyse de Rasch permet de mesurer la fonction du MS en classant les éléments selon un continuum de difficultés, puis de noter les patients selon un continuum d'aptitudes.

L'objectif global de cette thèse était de développer une mesure du MS spécifiquement conçue pour les enfants et les jeunes atteints d'AMC en utilisant l'engagement des parties prenantes et l'analyse de Rasch. Pour concrétiser l'objectif général de la thèse de doctorat, quatre études de recherche ont été menées et constituent les trois manuscrits de cette thèse.

La première étude était une revue de la littérature (Manuscrit 1) visant à identifier les évaluations basées sur la performance les plus fréquemment utilisées pour évaluer la fonction du MS dans la réadaptation pédiatrique. Le deuxième objectif était de déterminer le lien entre les concepts du « International Classification of Function, Disability, and Health (ICF) » et les concepts extraits de chaque mesure. Les résultats ont montré que les domaines de la participation et des facteurs personnels étaient mal représentés. Les résultats de cette revue ont renforcé la nécessité d'impliquer les parties prenantes dans la prochaine phase de la thèse.

Les deuxième et troisième études (Manuscrit 2) visaient à déterminer les éléments les plus utiles d'un point de vue clinique à inclure dans une évaluation pour les enfants atteints d'AMC.

Pour ce faire, une technique de groupe nominal a été utilisée avec un adulte atteint d'AMC et des parents, ainsi qu'un sondage clinique à trois volets. La version préliminaire du « Shriners Hospitals Arthrogryposis Pediatric Evaluation - UPper extremity (SHAPE-UP) » a été développé baser sur l'engagement des parties prenantes.

L'étude finale (Manuscrit 3) consistait à administrer le SHAPE-UP à 92 participants répartis sur six sites. Les objectifs de cette étude étaient 1) d'évaluer les propriétés psychométriques du SHAPE-UP en utilisant l'analyse de Rasch et 2) de déterminer la force de la corrélation entre le nombre d'articulations impliquées et le score finale. L'échelle de « Task Completion » du SHAPE-UP a été modifiée pour adopter une échelle de notation en 3 points. L'évaluation psychométrique initiale utilisant l'analyse de Rasch a fourni des preuves de la validité du contenu et de la construction ainsi que de la fiabilité.

La contribution originale de cette recherche doctorale est le développement de la première mesure pédiatrique du MS spécifique à l'AMC, le SHAPE-UP informé par les jeunes, les familles d'enfants atteints d'AMC et l'opinion d'experts cliniciens.

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Contribution to Original Knowledge

This thesis consists of original material that has not been published elsewhere, except where explicitly referenced. The research presented contributes to the advancement of knowledge in the field of pediatric rehabilitation and measurement.

This work will contribute knowledge towards evidence-based interventions to guide the treatment and evaluation of children and youth with Arthrogryposis Multiplex Congenita (AMC). The deliverable of the doctoral thesis is the Shriners Hospital Arthrogryposis Pediatric Evaluation - UPper Extremity measure, or SHAPE-UP, the first AMC-specific upper extremity (UE) outcome measure. This multicenter project will deliver significant contribution to the field of AMC as it will provide a novel evidence-based outcome measure in order to guide and inform clinical care. It is also expected to elevate the field at both the clinical and research levels. Evaluating the outcomes of treatment can inform the timing of specific interventions, which then facilitates the evaluation of treatment effectiveness. Outcomes of surgical and clinical (e.g., bracing, casting) interventions on large samples can be evaluated prospectively, rather than retrospectively. This data will inform clinicians and researchers as to the effectiveness and suitable timing of specific interventions to improve functional outcomes, minimize morbidity, and promote quality of life, thus fostering treatment advances in the field. Specifically, improved health care services allocation and cost-effectiveness of interventions can be anticipated.

All original data presented in this thesis was collected primarily at Shriners Hospitals for Children – Canada. Data from the fourth study, which entailed administration of the SHAPE-UP to children 1-21 years of age with AMC was collected from six participating sites; Shriners Hospitals for Children in Canada, Chicago, Greenville, Northern California, Philadelphia, and Portland. The Ethics Board of McGill University and local ethics board for the five other participating sites in the United States approved the studies involving research participants.

Contribution of Authors

Prior to completing this manuscript-based thesis, a thesis protocol was prepared by Caroline Elfassy and approved by the thesis protocol defence committee members, Isabelle Gélinas, PhD, and Anouk Lamontagne, PhD, and by the thesis supervisors, Laurie Snider, PhD, and Noémi Dahan-Oliel, PhD.

The manuscripts in this thesis are the work of doctoral candidate Caroline Elfassy with guidance from doctoral supervisors (Drs. Laurie Snider and Noémi Dahan-Oliel).

Caroline Elfassy conducted the studies for all manuscripts including the study design, statistical analyses, interpretation of outcomes and manuscript writing.

Drs. Laurie Snider and Noémi Dahan-Oliel oversaw all aspects of the thesis and provided expertise and guidance in research methodology.

Drs. Johanne Higgins and Lisa Wagner aided in the statistical analysis (Rasch) and outcome measure development.

Sarah Cachecho, Tessah Dunn, Clarice Araujo, and Rose Elekanachi were co-authors on the first manuscript for their role in data acquisition and analysis and for providing editorial feedback.

Kathleen Montpetit helped with the survey, content regarding the case report forms and provided guidance for the ethics component.

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Preface

My affinity for clinical research began during my Master's degree in Occupational Therapy (OT) at McGill University. The goal of our Masters group project was to investigate burn survivors' and clinicians' perspectives of the barriers and facilitators to work reintegration. After completing this project, I was able to witness firsthand the positive impact of clinical research and knowledge dissemination based on gaps identified by key stakeholders. Additionally, I've always had a particular interest for hand therapy, specifically fabricating orthoses. This led me to pursue an occupational therapist position in conjunction with a role as a research assistant at Shriners Hospitals for Children – Canada. When I started working at Shriners, a new multidisciplinary clinic was implemented to evaluate and treat children with a rare condition, Arthrogryposis Multiplex Congenita (AMC). Seeing as there was a need for an OT at the aforementioned clinic, it would become my role to provide evidence-based care to this population. The literature was sparse regarding rehabilitation guidelines or interventions for children with AMC. Therefore, the clinical research department in collaboration with the rehabilitation department decided to identify the needs surrounding rehabilitation as experienced by youth with AMC, caregivers, and clinicians to propose solutions to develop family- and client-centered rehabilitation recommendations. Based on the information gathered and through the feedback shared by the clinicians, it became evident that a condition-specific outcome-measure was to be developed.

Just like during my Master's project, acknowledging the gap in literature was one piece of the puzzle, but it remained crucial to address this gap in order to provide the best possible care to a population that was under-represented in the literature. Therefore, I was offered the opportunity to work collaboratively in a large multisite study to develop an upper extremity outcome measure specific to children and youth with AMC. After collaborating with multiple stakeholders and participants, I am pleased to present this doctoral thesis.

Thesis Organization and Overview

This thesis is manuscript-based and prepared in accordance with the regulations outlined by the Graduate and Postdoctoral Studies (GPS) Office. The thesis consists of a collection of three original manuscripts. The first manuscript was accepted to the Canadian Journal of Occupational Therapy and is currently In Press. The second manuscript was published in Research Involvement and Engagement. The third manuscript is submitted to Archives of Physical Medicine and Rehabilitation. As per the guidelines set by the GPS, a literature review and conclusion separate from the manuscripts as well as additional chapters have been included in this thesis. Each manuscript represents a sequential phase aiming towards the completion of this doctoral project. Therefore, duplication of the material is inevitable.

The thesis is organized in eight chapters:

Chapters 1 and 2 provide an introduction and a literature review on pediatric conditions and rare diseases, AMC, measurement in pediatric rehabilitation, the construct of UE, as well as rehabilitation related to AMC.

Chapter 3 explains the rationale, overall aim and specific aims of the thesis.

Chapter 4 presents the methodological basis of this thesis.

Chapter 5 consists of the first manuscript entitled "*Pediatric performance-based outcome measures for upper extremity function: Scoping review*" submitted to the Canadian Journal of Occupational Therapy. This scoping review reports the various performance-based pediatric outcome measures used clinically as well as providing an in-depth analysis on linking extracted outcome measure items to the ICF.

Chapter 6 consists of the second manuscript entitled *"Stakeholder Engagement in the Development of an Upper Extremity Outcome Measure for Children with Rare Musculoskeletal Conditions"* published in Research Involvement Engagement. With stakeholder engagement at the center, the objectives were to determine the most clinically useful items to be included in an AMC-specific UE function outcome measure.

Chapter 7 consists of the third manuscript entitled "*Initial psychometric evaluation of an upper extremity Arthrogryposis Multiplex Congenita-specific measure using Rasch analysis: A cross-sectional study*" submitted to Archives of Physical Medicine and Rehabilitation. The objectives were to evaluate the psychometric properties of the SHAPE-UP Task Completion scale using

Rasch analysis and 2) determine the strength of the correlation between the number of joints involved and the Task Completion score.

Chapter 8 involves a summary of the main findings, a general discussion, strengths and limitations, implications for future research and a conclusion.

An overall bibliography for all other chapters is presented at the end of this thesis.

Each manuscript contains its own set of tables, figures, references and appendices. Given the manuscript-style formal of this thesis, the inclusion of extensive tables illustrating the ICF linking process in Manuscript 1 (Appendix 1 p.118 and 2 p.173) and the supplementary files displaying the 3-round surveys in Manuscript 2 (Appendix, 3 p.190, Appendix 4 p.198, and Appendix 5 p.216) have been strategically placed in the Appendices section. This formatting ensures a smoother narrative for the reader.

Chapter 1: Review of the Literature

1.1 Overview of Pediatric Conditions

The World Health Organization (WHO) estimates the number of children aged 0-14 years experiencing "moderate or severe disability" at 93 million (5.1%), with 13 million (0.7%) children experiencing severe difficulties [1,2]. The United Nations Children's Fund estimated in 2005 that the number of children with disabilities under the age of 18 years at 150 million [3]. The variation in prevalence may largely be due to underrepresentation and misrepresentation of children with disabilities worldwide. In 2006, Statistics Canada created the Participation and Activity Limitation Survey (PALS), a national survey, to determine the number of Canadian children and adults with an activity limitation, the type of limitation experienced, and barriers they might face due to their health-related condition. Approximately 3.7% of Canadian children under the age of 15 were reported as having one or more activity limitation [4]. Population data demonstrate that musculoskeletal (MSK) conditions are among the most prevalent types of chronic conditions worldwide, and are a significant cause of pain and disability [5–7]. MSK conditions comprise a wide spectrum of aetiologies, including congenital or acquired injury to the bones, soft-tissue and joints [8]. Additionally, MSK conditions are associated with elevated costs to society, as children and adolescents under the age of 20 years with MSK deformities cost the United States $7.7 million in 2012 [9]. As with most chronic pediatric conditions, the indirect burden of pediatric MSK conditions is amplified by the effect on the family and caregivers. The emotional impact that chronic MSK conditions have on the family is immeasurable [9]. In contrast to adult conditions, pediatric MSK conditions may have lifelong ramifications resulting in the compounding of burdens over time [9]. Indeed, the PALS study identified that 16.8% of parents of children with a mild to moderate disability reported that their main source of stress was their child's health, whereas more than twice as many (38.3%) parents of children with a severe to very severe disability stated the same [4].

1.2 Overview of Pediatric Rare Diseases

While there is no universal definition of rare disease, the concept is oftentimes linked based on a prevalence threshold [10,11]. A recent study [10] published in the *European Journal of Human Genetics* used epidemiological data in the Orphanet database to calculate the global point prevalence estimate of rare diseases. They stated that approximately 4% of the total world

population is affected by a rare disease at any given time [10]. In America, rare diseases are defined as disorders affecting fewer than 200 000 people in the country or 86 per 100 000 people. Other definitions range in prevalence from 5 to 76 per 100 000 [10,12,13]. Therefore, like with MSK conditions, the true number of children living with a rare disease is unknown.

Pediatric rare diseases are often complex and chronic with limited treatment options [12]. Evidence on effectiveness of interventions are challenging to implement due to small samples, geographically dispersed, and heterogeneous populations [14]. Parents and children are unable to make informed decisions regarding treatments leading to difficult disease management [12,14]. Currently, condition specific outcome measures that are relevant to patients with rare diseases are extremely limited [15]. One such group of pediatric rare MSK conditions that has limited information regarding treatment management, which affects the child's activity and participation, is arthrogryposis multiplex congenita (AMC).

1.3 Arthrogryposis Multiplex Congenita

1.3.1 Etiology

The term AMC is derived from the Greek words "arthro" meaning joint, "gryposis" meaning curved, "multiplex" meaning that multiple joints are involved, and "congenita" referring to the contractures being present at birth (i.e., congenital). "Arthrogryposis multiplex congenita" was first used by Dr. Walter G. Stern, an orthopedic surgeon, during a presentation to the American Medical Association in June 1923, then published in JAMA in November 1923 [16]. Since then, the terms arthrogryposis, AMC, and multiple congenital contractures, have been used individually or interchangeably in hundreds of articles to describe the common feature of multiple contractures present at birth. Over the years, the terminology used when defining AMC has evolved. In the earlier literature, AMC was described as a diagnosis for a child born with multiple congenital contractures [17–21]. The words arthrogryposis and Amyoplasia have also been used interchangeably in the literature [22–24]. However, in the past 40 years, it has become widely accepted that Amyoplasia is a specific diagnosis, whereas arthrogryposis and AMC are broader interchangeable terms describing a group of lifelong conditions of varying etiology that limit joint movement [25–31]. For purposes of consistency, the term AMC will be used throughout this thesis. Over 400 conditions have been identified as having multiple congenital contractures as a clinical sign [32]. These conditions vary widely in terms of genetic origin, pathophysiology, and clinical

presentation, which makes it evident that AMC should be considered to be a descriptive term and not a specific diagnosis. Therefore, an international team of experts in which I had the opportunity to contribute [33,34] developed a consensus-based AMC definition in 2019, which reads as follows:

> Arthrogryposis multiplex congenita (AMC) is a term used to describe a group of congenital conditions characterized by joint contractures in two or more body areas. While the precise cause may be unknown for some individuals, causes are variable and may include genetic, parental, and environmental factors, as well as abnormalities during fetal development. Individuals with AMC have limited joint movement, with or without muscle weakness, in the involved body areas. Contractures vary in distribution and severity, do not progress to previously unaffected joints, but may change over time due to growth and treatment. Spinal deformities may be present at birth or develop throughout childhood and adolescence. Depending on the underlying diagnosis, other body systems such as the central nervous system (CNS), respiratory, gastrointestinal, and genitourinary systems may be affected. Cognition may be affected if the CNS is involved; sensation is usually intact. The impact on mobility, activities of daily living, and participation is variable.

1.3.2 Prevalence and classification

Over the years, there have been many classification systems proposed but the two most popular classification systems are *Hall* [35] and *Bamshad* [36]. Hall's classification system is based on clinical evaluation and can be divided into three main groups of disorders: 1) primarily musculoskeletal involvement, 2) musculoskeletal involvement plus other system anomalies, and 3) musculoskeletal plus central nervous system involvement [35]. On the other hand, Bamshad's classification is primarily based on etiology and is also separated into three groups: 1) Amyoplasia, 2) Distal Arthrogryposis (further divided into 10 subtypes of distal arthrogryposis syndromes), and 3) Syndromic (divided into central nervous system etiology or progressive neurological etiology) [36]. Although both classification systems offer a better understanding of clinical presentation, they do not describe the full spectrum of phenotype or underlying diagnosis. It would be important to expand beyond the MSK system and consider the involvement of other systems such as the gastrointestinal, genito-urinary, and the central nervous systems. Seeing as AMC is such a heterogenous condition and a classification system does not currently exist, determining functional

ability in a standardized fashion, particularly of the UE, might allow clinicians to be able to better adopt a common set of ideas and terminology about this condition.

1.3.3 Diagnosis

In order to make a specific diagnosis, careful evaluation is required for the following information: a full 3-generation family history, a detailed physical examination with documentation of which parts of the body are involved, the degree of flexion or extension of various joints, photographs at different ages and detailed measurements (including the range of motion of various joints) [32,35]. Hall (2014) further describes in detail the clinical evaluation (e.g. pregnancy history, family history, newborn evaluation, malformations, response to therapy and changes with time) to be completed in order to reach an AMC diagnosis. In addition to the history and clinical evaluation, a laboratory evaluation may be performed to rule out specific causes of arthrogryposis (e.g. neuropathic versus myopathies) [32].

1.4 The Field of Measurement in Pediatric Rehabilitation

In the field of rehabilitation, specifically pediatric rehabilitation, measuring function for clinical assessment is essential to developing a treatment plan and ensuring the patient achieves their goals. Outcome measurements are central to clinical practice, and medical and health research as it informs and guides diagnosis, prognosis, and evaluation of the effectiveness of clinical interventions. For almost every population group, there are number of instruments or outcome measures that can be used to measure clinical conditions or health status. There exist generic types of outcome measures and disease-specific types of outcome measures. Generic measures are necessary to compare outcomes across different populations and interventions, particularly for cost-effectiveness studies. Disease-specific measures assess the special states and concerns of diagnostic groups [37]. Specific measures may be more sensitive for the detection and quantification of small changes that are important to clinicians or patients [38]. However, one of the major challenges faced by clinicians and rehabilitation professionals is ensuring that the measures used in clinical practice are psychometrically sound.

1.4.1 Psychometric paradigms in measurement

Three main psychometric paradigms (i.e., Classical Test Theory (CTT) [39], Item Response Theory (IRT) [40], and Rasch Measurement Theory (RMT) [41]) presently exist. These theories enable the prediction of outcomes measures by identifying parameters of item difficulty and the ability of test takers. A brief overview will be presented for each psychometric paradigm as well as the rationale for using RMT in this thesis.

1.4.1.1 Classical Test Theory (CTT)

Most of the outcome measures used in rehabilitation were developed using CTT. CTT can be traced back to Spearman who introduced the decomposition of an observed score into a true score and an error and estimated the reliability of observed scores [42]. CTT assumes that each individual has a true score which would be obtained if there were no errors in measurement. Error is often assumed to be a random variable having a normal distribution. The score achieved by an individual is rarely the individual's true score. This means that the true score will not change with repeated administration of the same test and that the score of a particular examinee depends on the particular test (i.e., the particular items that comprise that test) [43,44].

1.4.1.2 Item Response Theory (IRT)

IRT refers to a family of mathematical models that attempt to explain the relationship between latent traits (unobservable characteristics or attributes) and their manifestations (observed outcomes). IRT assumes that the latent construct and items of a measure are organized in an unobservable continuum with the main purpose focusing on establishing the individual's position on that continuum [40]. In this approach, each item on a test has its own item characteristic curve that describes the probability of getting each particular item right or wrong given the ability of the examinee. As seen in Figure 1-1, the item difficulty parameter corresponds to the location on the ability axis at which the probability of a correct response is 0.50. It is shown in the curve that item 1 is easier and item 2 and 3 have the same difficulty at 0.50 probability of correct response.

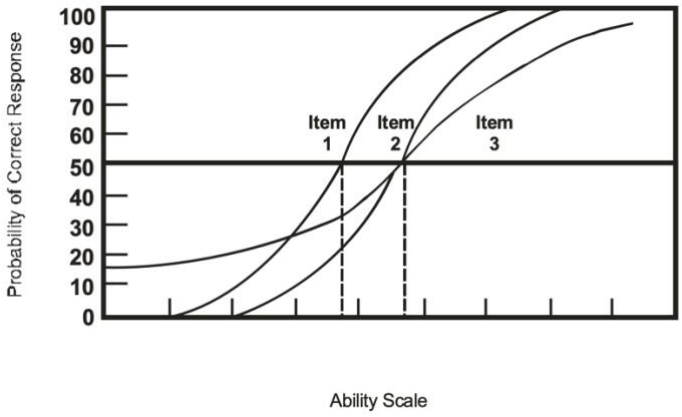

Figure 1-1. Hypothetical Item Characteristic Curves for Three Items using a Three Parameter Model

1.4.1.3 Rasch Measurement Theory (RMT)

RMT was born out of the work of Danish mathematician Georg Rasch. He developed the simple logistic model and through applications in education and psychology, he argued that he was able to demonstrate, mathematically, that his approach met stringent criteria for measurement [41]. The Rasch model is built on the assumption that the most parsimonious and effective predictor of a trait is the relationship between the difficulty of an item and the ability of a person [41]. The Rasch model exists independently of the data and could form an external criterion that the data could be tested against [45].

1.4.2 Limitations of CTT

There exist three major theoretical limitations to CTT: 1) the difficulty of the items is dependent on the ability of the group of persons to whom the items were administered, 2) the scales developed produce ordinal scores, where the difference between two adjacent scores at different points on the scale may not be equal, 3) the performance of scales is dependent on the particular sample in which they are used [44,46,47].

Modern psychometric methods such as IRT and Rasch can overcome these limitations as both can transform ordinal scales into interval measures. However, the main difference is that IRT models are descriptive and required a model-to-data fit, while the Rasch model was presented as a prescriptive model that demands a data-to-model fit [48,49].

1.4.3 Conceptual models and outcome measure development

According to Henrica de Vet (2011), test developers should model their newly developed outcome measures on conceptual models to help generate content to be measured [50–53]. This infers that conceptual models offer a structure for organization of subcomponents and assist in identifying necessary concepts to be included in newly developed outcome measures [54]. The three most frequently used health related quality of life (HRQOL) conceptual model found in the literature are the ones from Wilson and Cleary, Ferrans and colleagues, and the World Health Organization (WHO) [55]. A brief description of each conceptual model will be presented below.

Wilson and Cleary's [56] model of HRQOL combines two paradigms, biomedical and social science. Their model (Figure 1-2) is a taxonomy that includes five major well-defined domains: biological, symptoms, function, general health perception, and overall HRQOL. However, the definitions for two other domains, individual and environmental characteristics, were not made explicit. Each domain is related to the others, and reciprocal relationships may exists [55]. Wilson and Cleary suggest that environmental and individual factors are associated with outcomes, thus affecting total HRQOL [56].

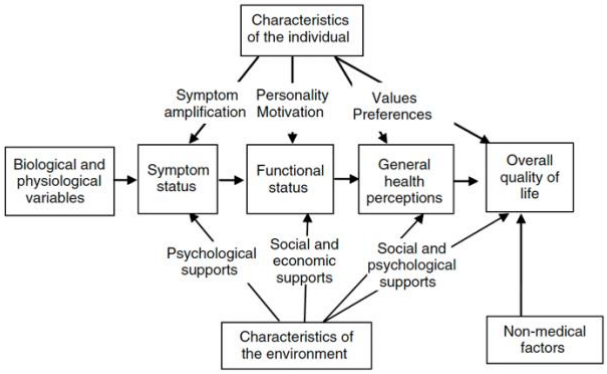

Figure 1-2 Wilson and Cleary (1995) HRQOL Conceptual Model

Ferrans, Zerwic, Wilbur, and Larson published a revised version of the Wilson and Cleary model [57] (Figure 1-3). The five major domains of the original model were kept; however, explicit definitions for individual and environmental characteristics were developed. Additionally, the depiction of the model was simplified by removing non-medical factors and labels on the arrows portraying the relationships in the figure. The model depicts dominant causal associations with implied reciprocal relationships [57]. The revised model can be applied to any healthcare discipline [55].

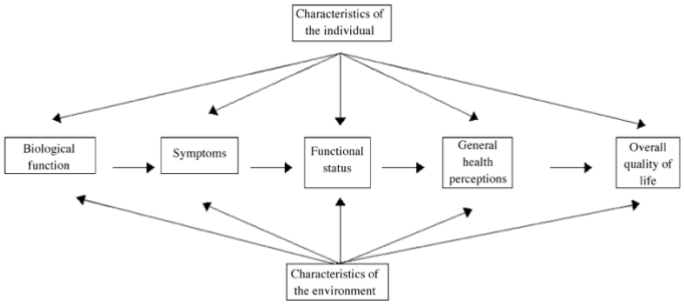

Figure 1-3 Ferrans et al. (2005) Revised version of the Wilson and Cleary model

In 2001, the WHO published the International Classification of Functioning, Disability, and Health (ICF) in order to provide a framework and classification system to measure health care needs based on a common language and definition of key words [58]. They stated that "health does not solely rely on physical function but rather is influenced by a multitude of factors including psychosocial well-being, participation and the interplay with the individual's environment" [59]. In 2007, the WHO developed a pediatric-specific version of the ICF, the International Classification of Functioning, Disability, and Health – Child and Youth (ICF-CY), in order to address four key issues: the child in the context of family, developmental delay, participation, and environments [60,61]. In 2012, the WHO Family International Classification Council advised that the ICF-CY merge with the ICF and the additional items be added to enhance the ICF [62]. See Figure 1-4 for a diagram of the ICF Framework.

The ICF organizes information in two parts: functioning and disability and contextual factors. There are three domains of functioning (i.e. Body Functions and Structures, Activity, and Participation) which are associated with respective domains of disability (i.e. impairments, limitations, and restrictions). The contextual factors are organized according to the Environmental and Personal Factors. Based on the ICF Framework, Body Functions and Structures can be defined as physiological function as well as anatomical parts of the body and its systems. Activities is defined as the execution of tasks or actions by an individual (e.g., mobility, self-care, communication, school related tasks). Participation refers to involvement in a life situation (e.g. community, social, civic activity). This can include participation in leisure activities such as sports or music, participation in paid or unpaid work, and social relationships [58,61]. Participation can be influenced by the individual's environment, personal factors, and by health factors. In childhood, participation in different life roles, at home, school or in the community, is important as it contributes to the child's health and development. It allows the child to develop new

competencies and skills, promotes self-confidence, and helps with developing new relationships [63].

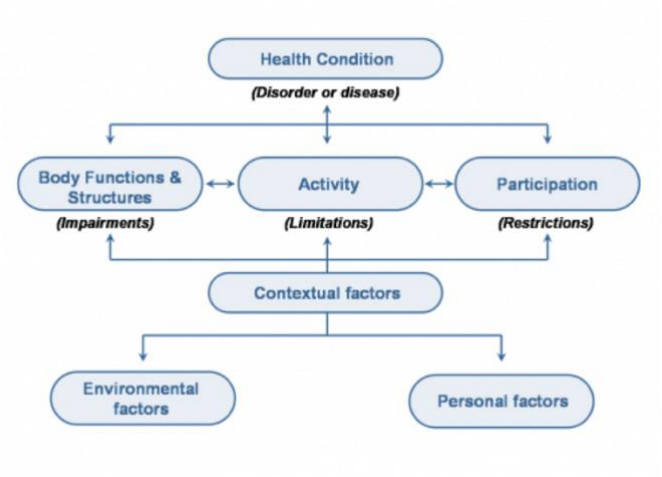

Figure 1-4 ICF Framework

When developing an outcome measure for a pediatric population with diverse clinical presentations, it is crucial to consider why the ICF framework is preferred over models like Wilson and Cleary and Ferrans and colleagues' models. Unlike these models, the ICF is not solely focused on HRQOL. Instead it serves as a comprehensive mapping and classification framework related to functioning as well as disability. This makes the ICF framework particularly suitable for accommodating the heterogeneous needs of pediatric populations. Moreover, while models like the Wilson and Cleary and Ferrans and colleagues' models may require adaptations to be used effectively with individuals unable to report their own HRQOL, such as infants, young children, and those with cognitive impairments [55], the ICF provides a more inclusive approach. It allows for a broader range of assessments making it applicable across various practice situations for needs assessments, treatment matching with conditions, and outcome evaluation.

1.5 Defining the Construct of Upper Extremity Function

The noun "function" has multiple definitions: 1) professional or official position, 2) the action for which a person or thing is specially fitted or used or for which a thing exists, 3) any of a group of related actions contributing to a larger action, especially the normal and specific contribution of a bodily part to the economy of a living organism [64]. Function in the context of health and illness has been well explored in the literature. In rehabilitation, an individual's ability to function is the basis for practice and directs clinical decision-making [65,66]. Determining a

patient's functional status through formal assessment is important for treatment planning and measuring rehabilitation outcomes [50]. Unsworth (1993) describes that the term function is explored and attempted to be defined as an abstract concept, yet there remained a lack of clarity in the literature concerning function. They conclude that more theoretical work is required to ensure that theory drives the measurement of function [67].

1.6 The Impact of AMC on Upper Extremity Function: Impairments, Limitations, and Restrictions

In regards to AMC, there have been reports in the literature that all spheres of the ICF framework are impacted due to this condition [68–70]. Specifically, children and youth with AMC will almost always have decreased strength and range of motion in two or more joints [71]. In terms of the impact on activities, children with AMC were reported to have activity limitations such as eating, bathing, toileting, and dressing as compared to typically developing peers [68,69]. A review specific to the participation of children and youth with AMC also demonstrated the paucity of information regarding participation such as sporting activities, employment, and education with this population [70]. Indeed, the rehabilitation process assists the individual with AMC to achieve their full potential by focusing on three levels of treatment, body functions and structures, activity, and participation.

1.7 Rehabilitation across the Lifespan in AMC

In order to optimize the quality of life of a child with AMC, rehabilitation and treatment interventions should start early, be comprehensive, and multidisciplinary [71,72]. The overall goals of rehabilitation are to facilitate and promote maximal independent function in activities of daily living and address participation in life roles [71]. Goals and objectives to treatment differ and evolve based upon developmental stages (i.e., infant, toddler, school-age, and teenage/adult), [69]. For example, a child in the infant stage will have more frequent visits with the multidisciplinary team than a child in the school age stage. During the infant stage, exploratory play is of utmost important while in the school age stage focus on autonomy and maximizing independence takes precedence. Additionally, transition from youth to adulthood and psychosocial wellbeing need to be addressed in this population, as elicited during the 3rd International Symposium on Arthrogryposis held in Philadelphia, USA in September 2019 [73].

Although the objective of rehabilitation might differ throughout the years and across the lifespan, a multidisciplinary team for children with AMC has the potential to enhance patient care and promote quality of life. The multidisciplinary team should include pediatric physiatrists/pediatricians, pediatric orthopedic surgeons (upper and lower extremity specialists), geneticists, neurologists, occupational therapists and physical therapists, social workers, and hand certified therapists to meet each child's and youth's individual needs. By addressing the needs of children and their families accordingly, early initiation of rehabilitation can reduce acute health care costs and prevent disability in the long run [74].

In summary, there remains gaps in the literature surrounding AMC and particularly the field of rehabilitation within AMC such as determining the best type of interventions to improve UE function and rehabilitation guidelines to reduce activity limitations and participation restrictions. In terms of outcome measure development, developers must use a conceptual model as the basis and foundation to offer structure to the measure.

Chapter 2: Rationale and Objectives

2.1 Rationale of the thesis

Clinicians such as occupational therapists (OTs) and physical therapists (PTs) help individuals participate in meaningful activities and enable function in all environments through the therapeutic use of occupations, adaptive tools and techniques, and modification of environments [65,66,75]. The use of outcome measures can help clinicians evaluate a child's capacity and performance of a task which pertain to different domains of the ICF (i.e., body functions and structures, activities, and participation) and validate beneficial services. Capacity is described as what a person with a health condition can do in a standard environment while performance is defined as what they can actually do in their usual environment [76]. OTs and PTs use a variety of measurement and evaluation tools to assess a patient's level of functioning. Although many outcome measures exist, some were developed specifically to assess and evaluate a region of the body and others were tailored for specific pediatric populations [77]. As AMC is a rare condition, there currently does not exist any outcome measure to be used specifically for this population. A qualitative study of youth with AMC, their caregivers, and clinicians identified the need for an AMC-specific outcome measure [68]. Wall et al. reported that children with AMC with UE involvement reported lower UE function compared to population norms. They further stated that objective measurements of task performance can delineate limitation in UE function, yet no such outcome measure existed for this pediatric population [78]. Indeed, during the Second International Symposium on Arthrogryposis held in St-Petersburg, Russia in 2014 and the Third International Symposium on Arthrogryposis in Philadelphia in 2019 a need for the development of a standardized assessment of short- and long-term outcomes was identified [73,79].

Currently, clinicians utilize generic measures aimed at overall functional performance and/or motor function, such as the Bruininks-Oseretsky Second Edition (BOT-2), Functional Dexterity Test (FDT), Peabody Developmental Motor Scales Second Edition (PDMS-2), Disabilities of the Arm, Shoulder and Hand (DASH), Pediatric Evaluation of Disability Inventory (PEDI) and others. Although these standardized measures provide important information on the level of function of the child, their clinical application is limited as these measures do not: 1) apply to children with a hand deformity, 2) guide clinicians as to which specific joint or muscle limits functional performance, and 3) allow the use of normative data when adaptations are made to accommodate the limb deformities present in children with AMC. Furthermore, capturing the

method of task performance would be clinically useful as children with AMC often complete tasks differently, by using another body part (i.e., mouth or feet), adapted tools or a different strategy. Yet the existing outcome measures used currently fail to capture method of compensation and satisfaction with task performance.

The purpose for using measures can be divided into three categories: discrimination, prediction, and evaluation. The discriminative ability of a measure is important to ensure it can distinguish patient groups. The predictive measure is used to predict outcome or prognosis. The evaluative measure is useful in detecting magnitude of longitudinal change in an individual [50,80]. Additionally, guidelines for developing a comprehensive measure of UE function should contain a wide spectrum of items that span the complete range of item difficulty, from the easiest task accomplished by the most severely affected individual to the most difficult ones completed only by those with mild impairment [77]. The tasks to be included in the outcome measure should require both unilateral and bilateral activities as well as account for UE development based on appropriate age groups [77,81].

Children with AMC frequently have UE involvement, approximately 56% of children have upper and lower involvement and 17% have involvement of their upper extremities alone [82,83]. As children with AMC display variable clinical features often affecting all joints of the UE, the development of an UE AMC outcome measure would assist clinicians to better identify the joint that is most problematic and the impact of this body structure impairment at the activity and participation level. Particularly, by developing an evaluative outcome measure specific to children with AMC, clinicians would be able to assess patient changes over time thus guiding surgical and non-surgical treatment planning (i.e., conservative vs. surgical management). Furthermore, a standardized measure that provides reproducible scores on the performance of developmentally appropriate manual tasks is needed to understand the relationship between hand function, satisfaction with performance, and clinical phenotype among children with AMC, as well as facilitate discussion within the multidisciplinary team.

2.2 Objectives

Therefore, the overall objective of this thesis was to develop an outcome measure to evaluate the UE of children and youth with AMC. To achieve this goal, a series of studies were developed. It is important to note that each phase informed the subsequent one and will be presented as a separate manuscript for the clarity of this thesis, as follows:

1. To guide the development of the SHAPE-UP, the aims of the first study, a scoping review, were twofold. The first aim was to identify which performance-based outcome measures (PBOMs) are most frequently reported to evaluate upper extremity (UE) function in pediatric rehabilitation. The second aim was to determine the link between constructs of the International Classification of Functioning, Disability, and Health (ICF) and meaningful concepts extracted from each identified PBOM

   Manuscript 1: *Pediatric performance-based outcome measures for upper extremity function: Scoping review*
2. To ensure the perspectives and needs of caregivers of children with AMC and clinicians were considered in the development of the UE measure for AMC, a Nominal Group technique (NGT) with caregivers of children with AMC followed by a three-round survey with clinicians were carried out.

   Manuscript 2: *Stakeholder Engagement in the Development of an Upper Extremity Outcome Measure for Children with Rare Musculoskeletal Conditions*
3. In order to validate the outcome measure, the aims of this study were 1) to evaluate the psychometric properties of the Shiners Hospital Arthrogryposis Pediatric Evaluation – Upper Extremity (SHAPE-UP) Task Completion scale using Rasch analysis and 2) determine the strength of the correlation between the number of joints involved and the Task Completion score.

   Manuscript 3: *Initial psychometric evaluation of an upper extremity Arthrogryposis Multiplex Congenita-specific measure using Rasch Analysis: A cross-sectional study*

Chapter 3: Methodology

The development of the SHAPE-UP arose over four studies (Figure 3-1). Each study was used to guide the subsequent one and will be presented as such for the clarity of the thesis. A detailed description of the methodology of each study can be found within each corresponding manuscript (Chapters 4,5, and 6).

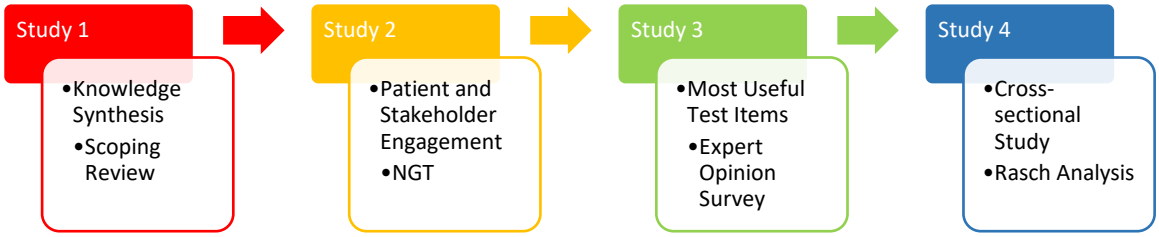

Figure 3-1 Development of the thesis

The first study, a scoping review, was conducted to identify all items related to pediatric UE performance-based outcome measure (PBOM). Items included in the review were mapped to the ICF. The first aim was to identify which PBOMs are most frequently reported to evaluate UE function in pediatric rehabilitation. The second aim was to determine the link between constructs of the ICF and meaningful concepts extracted from each identified PBOM. Refer to Chapter 4 for additional information.

Subsequently, Study 2 used a Nominal Group Technique (NGT) that invited youth with AMC and their family to determine the most important test items to consider during a clinical functional evaluation of the UE.

After completing the previous studies and identifying the relevant items specific to UE function based on the literature and patient-engagement priorities, Study 3 consisted of consultation with the experts in the field of AMC using a three-round survey to establish the different domains and associated items to be included in the outcome measure. Refer to Chapter 5 for additional information.

Finally, Study 4 entailed administering the newly developed SHAPE-UP outcome measure on a sample of children and youth with AMC to evaluate the psychometric properties using Rasch analysis and to determine the strength of correlation between joint involvement and Task Completion score. Refer to Chapter 6 for additional information

In summary, the four studies of the thesis were conducted to contribute to the development of the SHAPE-UP.

Chapter 4: Manuscript 1

4.1 Preface

A comprehensive scoping review was conducted to serve as a crucial starting point in the research process. This initial step involved examining the existing body of literature surrounding pediatric outcome measures, with a particular focus on performance-based assessments of upper extremity function. The scoping review had two aims: first, to identify which performance-based outcome measures (PBOMs) are most frequently reported to evaluate UE function in pediatric rehabilitation, and second, to determine the link between constructs of the ICF and meaningful concepts extracted from each identified PBOM.

Additionally, the scoping review uncovered significant insights into the existing gaps in the literature. These gaps were predominantly related to the domains within the ICF Framework that were not adequately covered by existing outcome measures. In essence, the review highlighted areas where future research and measurement development were necessary. These identified limitations not only informed the development AMC-specific outcome measure but also set the stage for subsequent chapters and suggest strategies for bridging these research gaps.

In summary, this scoping review was an essential first study that not only laid the foundation for the development of an AMC-specific outcome measure but also contributed valuable insights into the relationship between PBOM for UE and the ICF Framework.

4.2 Manuscript Title Page

TITLE: Pediatric performance-based outcome measures for upper extremity function: Scoping review

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Pediatric performance-based outcome measures for upper extremity function: Scoping review

Journal:	*Canadian Journal of Occupational Therapy*
Manuscript ID	CJOT-23-0047.R1
Manuscript Type:	Full research manuscript - quantitative/mixed methods
Mots-clés:	
Keywords:	Pediatrics*, Rehabilitation*, Outcome assessment (Health care)*, upper extremity, upper limb
Abstract:	Background: Performance-based outcome measures (PBOMs) are objective measures that assess physical capacity or performance in specific tasks or movements. Purpose: The first aim was to identify which PBOMs are most frequently reported to evaluate UE function in pediatric rehabilitation. The second aim was to determine the link between constructs of the ICF and meaningful concepts extracted from each identified PBOM Methods: Aim 1 consisted of a scoping review was performed. Pediatric UE PBOMs were searched in four electronic databases. The selection of UE outcome measures was conducted in two phases: an initial title and abstract screening, followed by full-text review of the articles to be included using selection criteria. Aim 2 consisted of linking items extracted from PBOMs to the ICF using the standardized Linking Rules developed by Cieza et al. (2019. Three reviewers analyzed the content and coded. Findings: After the initial screening, 1786 full-text articles were reviewed, 1191 met the inclusion criteria, in which 77 outcome measures were identified and 32 were included in the linking process. From the included 32 outcome measures, 538 items were extracted and linked to the ICF. Implications: A majority of the outcome measures identified were linked with the Chapter 4: Mobility, Fine hand use of the ICF. Therefore, when selecting a PBOM, careful considerations needs to be made regarding which concept of health is to be assessed.

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4.3 Abstract

Background: Performance-based outcome measures (PBOMs) are objective measures that assess physical capacity or performance in specific tasks or movements.

Purpose: The first aim was to identify which PBOMs are most frequently reported to evaluate upper extremity (UE) function in pediatric rehabilitation. The second aim was to determine the link between constructs of the International Classification of Functioning, Disability, and Health (ICF) and meaningful concepts extracted from each identified PBOM

Methods: Aim 1 consisted of conducting a scoping review. Pediatric UE PBOMs were searched in four electronic databases. The selection of UE outcome measures was conducted in two phases: an initial title and abstract screening, followed by full-text review of the articles to be included using selection criteria. Inclusion criteria comprised 1) be specific to the pediatric population (0-18 years) with any health status or health condition, 2) purpose of the PBOM included UE function, 3) outcome measure must be performance-based (PBOM), and 4) be published in English or French. Aim 2 consisted of linking items extracted from PBOMs to the ICF using the standardized Linking Rules developed by Cieza et al. (2019). Two reviewers were appointed to link the meaningful concepts identified in the outcome measures independently and a third reviewer was consulted in case of ambiguity to make a final decision.

Findings: After the initial screening, 1786 full-text articles were reviewed, 1191 met the inclusion criteria, in which 77 outcome measures were identified and 32 were included in the linking process. From the included 32 outcome measures, 538 items were extracted and linked to the ICF. The most commonly cited measures included the Assisting Hand Assessment, Jebsen-Taylor Hand Function Test, Melbourne Assessment of Unilateral Upper Limb, Quality of Upper Extremity Skills Test, the Box and Blocks. The Activity and Participation domain represented 364 codes (68%) followed by the Body Functions domain 174 codes (32%).

Implications: A majority of the outcome measures identified were linked with the Chapter 4: Mobility, Fine hand use of the ICF. Therefore, when selecting a PBOM, careful considerations needs to be made regarding which concept of health is to be assessed.

4.4 Introduction

Pediatric rehabilitation is a specialized field that aims to improve the functional outcomes of children with various health conditions (Hsu et al., 2021). Among the most common types of pediatric rehabilitation is musculoskeletal rehabilitation, which focuses on improving the physical function of children with bone, joint, and muscle disorders. In the field of rehabilitation, occupational therapists (OTs) and physical therapists (PTs) use various outcome measures to assess the effectiveness of their interventions (American Physical Therapy Association, 2014; Houtrow & Coster, 2019). These outcome measures can be either patient-reported or performance-based. Patient-reported outcome measures (PROMs) rely on self-reported information from the patient or caregiver, such as pain level or functional status. In contrast, performance-based outcome measures (PBOMs) require the patient to perform a set of movements or tasks that assess the patient's physical capacity or performance (Mayo, 2015). Scores for PBOMs can be based on either objective measurement (such as time to complete) or a qualitative assessment (such as describing task performance) that is assigned a score. PROMs and PBOMs can be either generic or condition-specific. Generic measures are designed to be applicable to a broad range of conditions and populations, while condition-specific measures are tailored to assess specific impairments or conditions for a given population. An example of a condition-specific measure for children with cerebral palsy would include the Gross Motor Performance Measure (GMPM) that seeks to evaluate gross motor performance or the Shriners Hospital Upper Extremity Evaluation (SHUEE) designed for use with hemiplegic type of cerebral palsy (Boyce et al., 1992; Davids et al., 2006).

While both types of outcome measures have their advantages and disadvantages, choosing the most appropriate measure for a specific patient and condition can be challenging. This is where the International Classification of Functioning, Disability and Health (ICF) can be a valuable framework for clinicians. The ICF provides a comprehensive and standardized system for describing health and health-related states, including body function and structure, activity, and participation (World Health Organization (WHO)., 2019). The ICF health-related states are further broken down into chapters (b=body function, s=body structure, d=activities and participation, e=environmental factors) followed by a numeric code. The number of digits which make up the code indicate the category and its level. The more digits (4^{th} level coding) available, the more precise description of the ICF (World Health Organization (WHO)., 2013). By using the ICF as a

framework to guide their choice of outcome measures, clinicians can ensure that they are assessing the most relevant and meaningful outcomes for their patients (Houtrow & Coster, 2019). This, in turn, can help better report the effectiveness of their interventions and ultimately improve the functional outcomes and quality of life of children with musculoskeletal conditions.

A recently published scoping review (Lesher et al., 2017) identified outcome measures frequently used in occupational therapy investigating the links between the constructs of the ICF and upper extremity outcome measures. Although the journal article by Lesher et al. (2017) provides an overview of linking constructs from UE outcome measures to the ICF, it is important to note that their approach did not follow a standardized linking process. Additionally, the authors were limited to first-level codes of the ICF, providing only a superficial level of information. A deeper analysis into the content of a measure can provide a comprehensive assessment of the most relevant components of a specified construct for a target audience and context (MacDermid, 2021). Hence, there was a need to conduct a more in-depth review of PBOMs across all pediatric conditions impacting the UE function in order to accurately inform clinicians in their selection of outcome measure to better evaluate children with UE anomalies.

In order to provide a standardized method for collecting health information, Alarcos Cieza and colleagues have proposed rules for linking existing health information to the ICF. Since its development in 2002, two updated versions have been published with the latest being in 2019 (Cieza et al. 2019). The latest version includes ten linking rules, as well as a linking extraction table. By implementing a standardized linking process, health-status measures can be properly used to assess and address patients' needs in rehabilitation.

The aims of this scoping review were twofold. The first aim was to identify which PBOMs are most frequently reported to evaluate UE function in pediatric rehabilitation. The second aim was to determine the link between constructs of the ICF and meaningful concepts extracted from each identified PBOM. The overall goal was to determine the gaps in the PBOMs to help better inform clinicians and outcome measure developers.

4.5 Methodology

Design:

A scoping review is a form of knowledge synthesis that addresses an exploratory research question aimed at mapping key concepts, types of evidence, and gaps in research related to a

defined area or field by systematically searching, selecting, and synthesizing existing knowledge (Arksey & O'Malley, 2005). The current scoping review used Arksey and O'Malley's (2005) framework in addition to the recommendations by Levac et al. (2010) (Arksey & O'Malley, 2005; Levac, 2010). This framework consists of five steps: i) identifying the research question(s), ii) identifying relevant studies, iii) selecting studies, iv) charting the data and v) collating, summarizing and reporting results (Arksey & O'Malley, 2005; Levac, 2010).

i) Identifying the research question

a. Literature Review

The first aim was to identify which PBOMs are most frequently reported to evaluate UE function in pediatric rehabilitation. UE function was defined as UE coordination, motor function, sensation, muscle strength, and stabilization.

b. Linking to the ICF

The second aim was to determine the link between constructs of the ICF and meaningful concepts extracted from each identified PBOM using standardized ICF Linking Rules (Cieza et al., 2019).

ii) Identifying relevant studies

a. Literature Review

A comprehensive search strategy was designed using the PRISMA extension for scoping reviews (PRISMA-ScR) (Tricco et al., 2018). In order to conduct a comprehensive search that would include UE evaluation for the pediatric population in rehabilitation, articles were searched through the following electronic databases: Medline (1950-2021), EMBASE (1980-2021), CINAHL (1990-2021), and OTseeker in July 2021. The databases were used to reach a broad-based search strategy of English and French literature. To identify relevant studies, members of the research team entered search terms that included but were not limited to "child","infant","adolescent","upper extremity","upper limb", "fine motor skills" "agility or function" in English in varying combinations into the electronic databases. The search strategy with associated MeSH terms and/or text word search or combinations for the search strategy in Medline can be found in table 1.

b. Linking to the ICF

Alarcos Cieza and colleagues have developed three iterations of linking rules, the latest being published in 2019. These were the linking rules applied during the linking process.

iii) Selecting studies

a. Literature Review

The selection of UE outcome measures (Aim 1) was conducted in two phases: an initial title and abstract screening, followed by full-text review of the articles to be included using the selection criteria. Two reviewers used the Rayyan web application, which allows an independent and blinded application of the selection criteria to the process of selecting studies for inclusion (Ouzzani et al., 2016), to go through the title and abstract screening portion. A third reviewer was included to resolve any disagreements in this initial selection process. To increase consistency among reviewers, all reviewers screened the same 50 publications and discussed the results before beginning screening. Once all reviewers agreed, the two reviewers then applied the selection criteria to full-text articles. For a study to be included, it had to meet the following conditions: 1) be specific to the pediatric population (0-18 years) with any health status or health condition, 2) purpose of the PBOM included UE function, 3) outcome measure must be performance-based, and 4) be published in English or French. If an outcome was a self-reported or proxy completed measure it was excluded. The selection criteria can be found in table 2.

b. Linking to the ICF

Two reviewers were appointed to link the meaningful concepts identified in the outcome measures independently and a third reviewer was consulted in case of ambiguity to make a final decision. A discussion was held to ensure consensus was reached. In addition to acquiring good knowledge of the conceptual and taxonomical fundamentals of the ICF, the three reviewers performed a mock linking session to align perspectives and ensure all had the same understanding of the linking rules. A kappa statistic was reported to determine the level of agreement for linking of meaningful concepts to the ICF. Kappa values generally range from 0 to 1 with 1 indicating perfect agreement and 0 indicating no additional agreement beyond what is expected chance alone. Kappa coefficients above 0.6 are regarded as good (Bernardelli et al., 2021). The data analysis was performed with SPSS.

iv) Charting the data

a. Literature Review

The included studies were organized in an excel document which identified the name of the outcome measure, number of times it was identified in the literature search, author and year of

publication, purpose, intended population, age of population, number of domain of function assessed, list of domains assessed (Table 3).

b. Linking to the ICF

The latest linking rules suggest using a specific linking extraction table that includes the following information: Name of outcome measure, verbatim health information, perspective adopted in information, response options, classification of response options, main meaningful concepts, additional meaningful concepts, ICF category for the main meaningful concepts, ICF category for the additional meaningful concepts, annotation (Cieza et al., 2019). This table was generated for each included outcome measure (Appendix 1).

v) Collating, summarizing and reporting results

Once the data from the measures was extracted and the items linked accordingly, the data was analyzed and synthesized. The outcome measures were organized based on the number of times they were reported in the literature. The items extracted from the outcome measures were then linked to the ICF following the linking extraction table (Appendix 1) (Cieza et al., 2019). Findings were reported and discussed below (Arksey & O'Malley, 2005).

4.6 Results

a. Literature Review

The search conducted in MEDLINE, EMBASE, CINAHL, and OTSeeker in July 2021 generated a total of 6775 articles. Duplicates were removed (n=802), and the selection criteria were applied to 5973 titles and abstracts. After the initial screening, 1786 full-text articles were reviewed, 1191 met the inclusion criteria, in which 77 outcome measures were identified and 32 were included in the linking process. A PRISMA flow chart was used to illustrate the identification and selection process of the articles and outcome measures at each stage of the review (Moher et al., 2009) (figure 1). The included outcome measures were published between 1948 and 2013. The most commonly cited included the Assisting Hand Assessment (AHA) (Krumlinde-Sundholm & Eliasson, 2003), Melbourne Assessment of Unilateral Upper Limb (MAUUL) (Randall et al., 1999), the Jebsen-Taylor Hand Function Test (JTHFT) (Jebsen et al., 1969), the Quality of Upper Extremity Skills Test (QUEST) (DeMatteo et al., 1992), the Box and Blocks (Mathiowetz et al., 1985). Refer to table 3 for an in-depth overview of data extracted from the included outcome measures.

b. Linking to the ICF

Five hundred and thirty-eight items were extracted from the 32 included outcome measures, which were then linked to the ICF. Some items were coded to more than one code as there were additional meaningful concepts found within the items. The kappa coefficient was calculated at 0.721 which indicates a substantial agreement among the three raters (McHugh, 2012). The Activity and Participation domain represented 364 codes (68%) followed by the Body Functions domain 174 codes (32%). The most frequently identified ICF codes were *d4402 – Manipulating (n=99, 18%) and d4401 – Grasping (n=85, 16%)*. The most coded ICF Chapter was *Chapter 4: Mobility*, specifically *Fine hand use* (n=301, 56%) and *Hand and arm use* (n=17%). As mentioned, a table for each outcome measure was created using the Cieza et al (2019) linking process (Appendix 1). Additionally, a pie chart was created for each outcome measure with the breakdown of codes identified (Appendix 2) (Figure 2. AHA as an example).

4.7 Discussion

a. Literature Review

This scoping review identified the most frequently reported pediatric PBOMs of UE function in the literature and linked the extracted items to the domains of the ICF. A total of 32 outcome measures that met the inclusion criteria were included and 538 items were linked to the ICF. The most frequently cited outcome measures included the AHA, the MAUUL, the JTHFT, the QUEST and the Box and Block. In terms of the ICF Linking process, the most frequently reported codes were *d4402 – Manipulating (n=99, 18%) and d4401 – Grasping (n=85, 16%)*.

A total of 27 of the included measures were developed specifically for a pediatric population, while the JTHFT, Box and Blocks, and FDT measures were originally developed for an adult population but were later validated for a pediatric population. The Purdue Pegboard Test and the Assessment of Capacity for Myoelectric Control (ACMC) can be used throughout the lifespan.

b. Linking to the ICF

One issue faced during the linking process was inherently related to the limited coding structure of the ICF itself. For example, the **ICF Framework** is divided into Health Condition, Body Functions and Structures, Activities, Participation, Environmental Factors, and Personal Factors (Figure 3a). However, the **ICF Coding Structure** is divided into Body Functions, Body

Structures, Activities and Participation, and Environmental Factors (Figure 3b). The major difference is the individual category of "Activity" and "Participation" in the ICF Framework while "Activities and Participation" is combined in the Coding Structure. By linking to the ICF Coding Structure, we were unable to determine which items were directly related to the Participation component and those related to Personal Factors could not be coded due to the nature of the coding structure. Therefore, although the domains of the ICF provided a helpful framework to understand the influence of various factors on the individual, the WHO's definition of participation provided little detail about the construct, making it difficult to use as a guide to select appropriate and specific measurement tools without a more refined parsing of the underlying concepts (Adair et al., 2018). Unfortunately, test developers aren't always aware of this limitation when developing new outcome measures. This could lead to items being duplicated or constructs being misrepresented and could further generate gaps between the constructs quantified by the outcome measure and those of the ICF when determining which ICF health-related components are lacking. As the items were coded to the Body Functions and Activity domains of the ICF, youth with conditions such as osteogenesis imperfecta (OI) and arthrogryposis multiplex congenita (AMC), who have reported experiencing difficulties with participation at home, at school, and in the wider community (Elfassy et al., 2019, 2020; Marr et al., 2017) will not be able to identify such needs using the ICF. When items are not coded to include the Participation domain of the ICF, it can result in an incomplete understanding of the individual's functioning and may lead to inadequate intervention strategies. Additionally, since Personal Factors are not coded in the ICF, this area is oftentimes overlooked. Therefore, it is important for clinicians administering outcome measures to carefully consider all domains when using the ICF to assess and classify individuals' functioning. Since treatment plans are often developed on the basis of the results of the assessments, clinicians should also consider the importance of participation for children and youth and the personal factors (Elfassy et al., 2019, 2020). This can be achieved through PBOMs but also through guided interviews using measures like the Canadian Occupational Performance Measure (COPM) and Goal Attainment Scaling (GAS) (Law et al., 2019; Palisano, 1993). OT and PTs working with children and youth should use their clinical judgment to ensure that a child and family-centered approach is implemented and that all needs are addressed.

As reported, the ICF is composed of four chapters (b=body function, s=body structure, d=activities and participation, e=environmental factors) followed by a numeric code. The number

of digits which make up the code indicate the category and its level. The more digits (4th level coding) available, the more precise description of the ICF (World Health Organization (WHO)., 2013). According to the linking rules "all meaningful concepts [should be linked] to the most precise ICF category" (Cieza et al., 2019). In the scoping review by Lesher et al (2017), the items were only linked to the first level category. For example, in the JTHFT, Lesher et al (2017) coded in Body Functions: b1"Mental" and b7 "Neuromuscular and Movement", in Activity and Participation: d1 "Learning and Applying Knowledge", d2 "General Tasks and Domains", d5 "Self-Care", d6 "Domestic Life", whereas the fourth level codes found in Table 4 were much more precise and specific. Linking the meaningful concepts to the most precise category is important for communication and collaboration between clinicians. When coding to the first level category of the ICF, granularity and precision are lost, which can ultimately result in a decreased understanding of the level of function of an individual. Precise coding helps to ensure that the different clinicians such as OTs, PTs and physicians are using the same language to describe an individual's functioning, which can facilitate communication and coordination of care (Selb et al., 2015; World Health Organization (WHO)., 2001).

Another important aspect to consider when linking the items of the outcome measures to the ICF was that some items were linked with more than one meaningful concept. Therefore, these items were linked to two or more codes. An example of an item with multiple meanings was found in the Performance of Upper Limb Test where the item stated "hand to mouth with and without weight". The meaningful concepts "hand to mouth", "with and without weight" were linked to *b7602 - Coordination of voluntary movements* and *b7301 - Power of muscles of one limb* respectively. This particular example is in line with Linking rule #3 which states "identify any additional concepts contained in the piece of information in addition to the main concepts already identified".

The second version of the linking rules (2005) had omitted the "other specified" and "unspecified" categories of the ICF to reduce the ambiguity of the results in the linking process (Cieza et al., 2005). However, in the latest version, Cieza et al. (2019) recommend the use of "other specified [8]" such as *picking up large heavy cans – d4408 Fine hand use, other specified* and "unspecified [9]" such as *proceeds – d4409 Fine hand use, unspecified* has shown that not using 8 and 9 categories led to lost information". The difference is that the content and information be added and written in the newly developed extraction table. The modification of this rule has been

necessary for this study as “other specified” represented 12% (n=65) of the results and “unspecified” denoted 3% (n=15). Coding to these two categories was beneficial because it allowed for the inclusion of meaningful concepts that did not fit precisely into other existing ICF categories. By using these categories, clinicians can still accurately describe the individual’s functioning and address all relevant aspects of their condition, even when no specific ICF category exists for a particular concept. This ensures that no meaningful aspect of the individual’s condition is omitted from the coding process (Cieza et al., 2019). Therefore, while the ICF is composed of four chapters and numeric codes, the more digits available for coding, the more precise and comprehensive the description of an individual’s functioning, which in turn can improve collaboration between clinicians and promote care.

4.8 Conclusion

The scoping review identified 32 PBOMs used in the assessment of pediatric UE function. The most frequented reported outcome measures were the AHA, JTHFT, MAAUL, QUEST, and Box and Blocks. A majority of the meaningful concepts extracted from the PBOMs were linked with the Chapter 4: Mobility, Fine hand use of the ICF. Due to the nature of the ICF Coding Structure, the areas of “Participation” and “Personal Factors” were lacking in the linking process. Overall, clinicians need to aware of the limitations between the ICF Framework and the ICF Coding structure and ensure they are addressing all the needs of children and youth. This can be achieved by combining assessments and outcome measures to bridge the gap between what is lacking in one measure and complement treatments provided to children and youth.

4.8.1 Limitations

In this latest iteration of Cieza et al.’s linking rules, the authors suggest using a Linking Extraction Table when linking health information to the ICF (Cieza et al., 2019). Specifically, they recommend extracting the “perspective adopted in information”, “response options”, and “classification of response options”. However, when linking PBOM, many categorizations of response options did not apply as the outcome measure was often related to function. Furthermore, since the outcome measures were to be performed in a clinical rehabilitation setting evaluated by a clinician, 28 out of the 32 measures were assessing capacity. Yet, contextual factors such as

physical and social environment, as well as personal factors such as motivation, influence the relations between capacity and performance (Holsbeeke et al., 2009).

4.9 Tables

Table 1. Search strategy in Medline (1947-July 2021)

#	Searches	Results
1	exp Child/	1993692
2	exp Infant/	1180056
3	Adolescent/	2111703
4	(newborn* or new-born* or neonat* or neo-nat* or infan* or child* or adolesc* or paediatr* or pediatr* or baby* or babies* or toddler* or kid or kids or boy* or girl* or juvenile* or teen* or youth* or pubescen* or preadolesc* or prepubesc* or preteen or tween).ti,ab.	2584896
5	exp Upper Extremity/ph, pp OR exp Hand Strength/ OR (exp Upper Extremity/ AND (exp "Musculoskeletal Physiological Phenomena"/ OR exp "Motor Skills"/ OR exp "Task Performance and Analysis"/ OR exp “Motor Skills Disorders” OR exp “Psychomotor Performace”))	67372
6	((hand* or arm* or elbow* or forearm* or shoulder* or manual or bimanual or upper extremit* or upper limb*) adj3 (activit* or abilit* or agility or function* or perform* or skill*)).ti,ab.	53084
7	5 or 6	109266
8	(Analy* or evaluat* or assessment* or assessing or instrument or instruments or measure or measurement* or measures or quantifying or scale or scales or score or scores or test or tests or tool or tools).ti.	2648737
9	1 or 2 or 3 or 4	4515271
10	7 and 8 and 9	2872

Table 2. Selection criteria to identify the performance-based outcome measures of UE for the pediatric population (Aim 1)

Inclusion criteria	Exclusion criteria
Pediatric population (0-18 years)	Adult population (>18 years)
UE function evaluated	PBOM not incorporating the evaluation of UE function
Performance-based	Self-reported; proxy-reported
English or French	No information on psychometric properties
Cited more than twice in the literature	
Access to a copy of the PBOM	

Table 3. Included outcome measures listed by most frequently reported in the literature.

Outcome measure name	Citation Count	Author (Year)	Purpose	Population intended	Age	# of Domains	Domains
Assisting Hand Assessment (AHA)	203	Krumlinde-Sundholm L., Eliasson A-C. (2003)	Measures and describes how children with an upper limb disability in one hand use his/her affected hand (assisting hand) collaboratively with the non-affected hand in bimanual play	Children with unilateral upper limb impairments	18 months-12 years	6	General usage, arm use items, grasp/release, fine motor adjustments, coordination, pace
Melbourne Assessment of Unilateral Upper Limb (MAUUL)	134	Randall M., Johnson L., Reddihough D. (1999)	Measure to quality of unilateral upper limb movement	Children with neurological conditions	2.5-15 years	14	NA
Jebsen-Taylor Hand Function Test	122	Jebsen, R. H., Taylor, N., Trieschmann, R. B., Trotter, M. J., & Howard, L. A. (1969)	Assess a broad aspects of hand function commonly used in activities of daily living	Neurological or musculoskeletal conditions	6 years and over	7	Writing, turning cards, picking up small common objects, simulated feeding, stacking checkers, picking up large light objects, picking up large heavy objects
Quality of Upper Extremity Skills Tests (QUEST)	105	DeMatteo, C., Law, M., Russell, D., Pollock, N., Rosenbaum, P., & Walter, S. (1992)	Evaluate quality of UE function	Children with neuromotor dysfunction with spasticity	18 months-8 years	4	Dissociated movement, grasp, protective extension, weight bearing
Box and Blocks	90	Hyres and Buhler (1957) & modified by Mathiowetz V, Volland G, Kashman N, Weber K (1985)	Evaluate unilateral gross manual dexterity	All children	3 years and over	1	Dominant and non-dominant hand
Bruininks-Oseretsky (BOT-2)	52	Bruininks, R., and Bruininks, B. (2005)	Measure fine and gross motor skills of children and youth	Children with motor impairments	4-21 years	4	Fine Manual control, manual coordination, body coordination, strength and agility
Movement Assessment Battery for Children (M-ABC)	51	Henderson, S. & Sugden, D. (1992)/ Henderson, S. E., Sugden, D. A., & Barnett, A. L. (2007).	Designed to identify and describe impairments in motor performance	All children	3-16	3	Manual Dexterity, Ball Skills, Static and Dynamic Balance
Functional Independence Measure for Children (WeeFIM)	40	Msall, ME., DiGaudia, K., Rogers, BT., LaForest, S., Catanzaro, NL., Campbell, J., Wilczenski, F., & Duffy, LC. (1994)	Measures the impact of developmental strengths and difficulties on independence at home, in school, and in the community	Children with disabilities	6 months-8 years	6	Self-care, sphincter control, mobility (transfers), locomotion, communication, and social cognition,
Peabody Developmental Motor Scales (PDMS)-2	32	M. Rhonda Folio M., & Fewell, R. (2000)	Provides an in-depth assessment and training or remediation of gross and fine motor skills	All children	Birth-5 years	6	Reflexes, stationary, locomotion, object manipulation, grasping, visual-motor integration
Performance of Upper Limb (PUL)	32	Mayhew A, Mazzone ESE, Eagle M, et al. (2013)	Assess upper limb function in ambulant and non-ambulant patients	Children with Duchenne muscular dystrophy	5-27 years	1	Upper limb function

Shriners Hospital for Children Upper Extremity Evaluation (SHUEE)	26	Davids JR, Peace LC, Wagner LV, Gidewall MA, Blackhurst DW, Roberson WM. (1996)	Assess the segmental, dynamic alignment of the involved UE when performing functional tasks	Children with hemiplegic cerebral palsy	3-18 years	6	AROM, PROM, spasticity, spontaneous functional analysis, dynamic positional analysis, grasp-and-release analysis
Purdue Pegboard Test	17	Tiffen & Asher (1948)	Measure unimanual and bimanual finger and hand dexterity	All children	5-89 years	2	Unimanual and bimanual dexterity
Motor Function Measure (MFM)	16	Bérard C, Payan C, Hodgkinson I, Fermanian J (2004)	Assess severity and disease progression of neuromuscular diseases	Children with neuromuscular diseases	Over 6 years	3	Standing and transfer, Axial and proximal motor function, Distal motor function
Functional Dexterity Test (FDT)	15	Aaron, D. & Jansem, C. (2003)	Measure the patient's ability to use the hand for functional daily tasks that require 3-jaw chuck prehension between the fingers and the thumb	Patient's with hand injuries	5 years and over	1	NA
Hand Assessment of Infants (HAI)	12	Krumlinde-Sundholm L, Ek L, Sicola E, Sjöstrand L, Guzzetta A, Sgandurra G, Cioni G, Eliasson AC. (2017)	Assess upper limb function	Infants at risk of developing cerebral palsy	3-12 months	2	Unimanual and bimanual
Pediatric Arm Function Test (PAFT)	10	Uswatte, G., Taub, E., Griffin, A., Rowe, J., Vogtle, L., & Barman, J. (2012)	Assess capacity to carry out actions and tasks with the more-affected arm	Children with hemiplegic cerebral palsy	2-8 years	2	Unilateral (proximal and distal) and bilateral
Upper Limb Physician Rating Scale (ULPRS)	10	Eun Sook Park, Ji-Woon Joo, Seon Ah Kim, Dong-Wook Rha, and Soo Jin Jung (2014)	Assesses movement quality of the upper limbs	Children with spastic cerebral palsy	5-13 years	1	NA
Assessment of Children Hand Skills (ACHS)	8	Chien, CW., Brown, T., McDonald, R. (2010)	Evaluate real life hand skill performance	Children with disabilities	2-12 years	3	Leisure and play, school/education, and activities of daily living
Test of Gross Motor Development (TGMD) -2	8	Ulrich (2000)	Assess gross motor functioning in children	preschool, early elementary, and special education	3-10 years	2	Locomotor and object control
Besta Scale	6	Fedrizzi, E., Pagliano, E., Andreucci, E., Oleari G. (2003)	Asses hand function	Children with hemiplegic cerebral palsy	18 months to 12 years	3	Grip, spontaneous use, stereognosis
Selective Control of the Upper Extremity Scale (SCUES)	6	Wagner, L.V, Davids, JR & Hardin JW (2016)	Assess selective motor control of the upper extremity in children	Children with unilateral cerebral palsy	3-18 years	1	Range of Motion
Bayley Scales of Infant and Toddler Development (BSID) - 3rd edition	5	Bayley (2006)	Assess the development of infants and toddlers	All children	1-42 months	5	Cognitive, Language, Motor, Social-Emotional, and Adaptive Behavior scales
Minnesota Handwriting Assessment	5	Reisman, J. (1999)	Analyzes handwriting skills with first and second grade students	All children	1st and 2nd grade (6-8 years)	5	Rate, legibility, form, alignment, size, spacing

Mini Assisting Hand Assessment (Mini AHA)	5	Greaves S, Imms C, Dodd K, Krumlinde-Sundholm L. (2013)	Measure how effectively children with unilateral cerebral palsy use their involved hand collaboratively with their well-functioning hand to perform bimanual play tasks.	Unilateral cerebral palsy	8-18 months	1	NA
Both Hands Assessment (BoHA)	5	Elvrum, A-K., Zethraeus, B-M., Vik, T., & Krumlinde-Sundholm, L.(2017)	Measure the effective use of both hands in bimanual activity performance, as well as to quantify the possible side difference between hands	Bilateral cerebral palsy	18 months – 12 years	1	NA
Unilateral Below Elbow Test (UBET)	5	Bagley, A., Molitor, F., Wagner, L., Tomhave, W., & James, M. (2006)	Measure how children use their prostheses to accomplish developmentally appropriate two-handed tasks, without their prosthesis and how the type prosthesis affects child's ability to accomplish functional tasks	Children with unilateral congenital below elbow deficiency	2-21 years	1	NA
Brachial Plexus Outcome Measure (BPOM)	4	Ho ES, Curtis CG, & Clarke HM (2012)	Measure the activity capacity of the affected limb	Children with obstetrical brachial plexus	4-19 years	1	NA
In-Hand Manipulation Test	4	Exner, C. (1990)	Assess the quality and efficiency of in-hand manipulation skills in children	All children	pre-school and school aged	1	NA
Assessment of Capacity for Myoelectric Control (ACMC)	4	Hermansson, L., Fisher, A., Bernspang, B., & Eliasson, A-C. (2005)	Assess the ability to control a myoelectric prosthetic hand	Upper limb reduction deficiency or amputation	Over 2 years	4	Gripping, holding, releasing, coordinating
Upper Extremity Rating Scale (UERS)	4	Koman LA, Williams RM, Evans PJ, et al. (2008)	Evaluate range of motion (a technical component of function)	Children with hemiplegic cerebral palsy	3-18 years	5	Shoulder active motion, elbow active motion, forearm active motion, wrist active motion, hand
Test of Infant Motor Performance (TIMP)	3	Campbell SK, Wright BD, Linacre JM. (2002)	Assessing the postural and selective control of movement needed for functional motor performance in early infancy.	Premature infants	34 weeks post-conception age to 4 months post-term	1	NA
Video Observations Aarts and Aarts, module Determine Developmental Disregard (VOA-DDD)	3	Aarts PB, Jongerius PH, Aarts, MA., Hartingsveldt, JV., Anderson, PG., & Beumer, A. (2007)	Extended evaluation of motor behaviour of affected upper limb	Children with spastic unilateral cerebral palsy	4-10 years	2	Duration and frequency of activity

4.10 Figures

Figure 1. PRISMA diagram of included outcome.

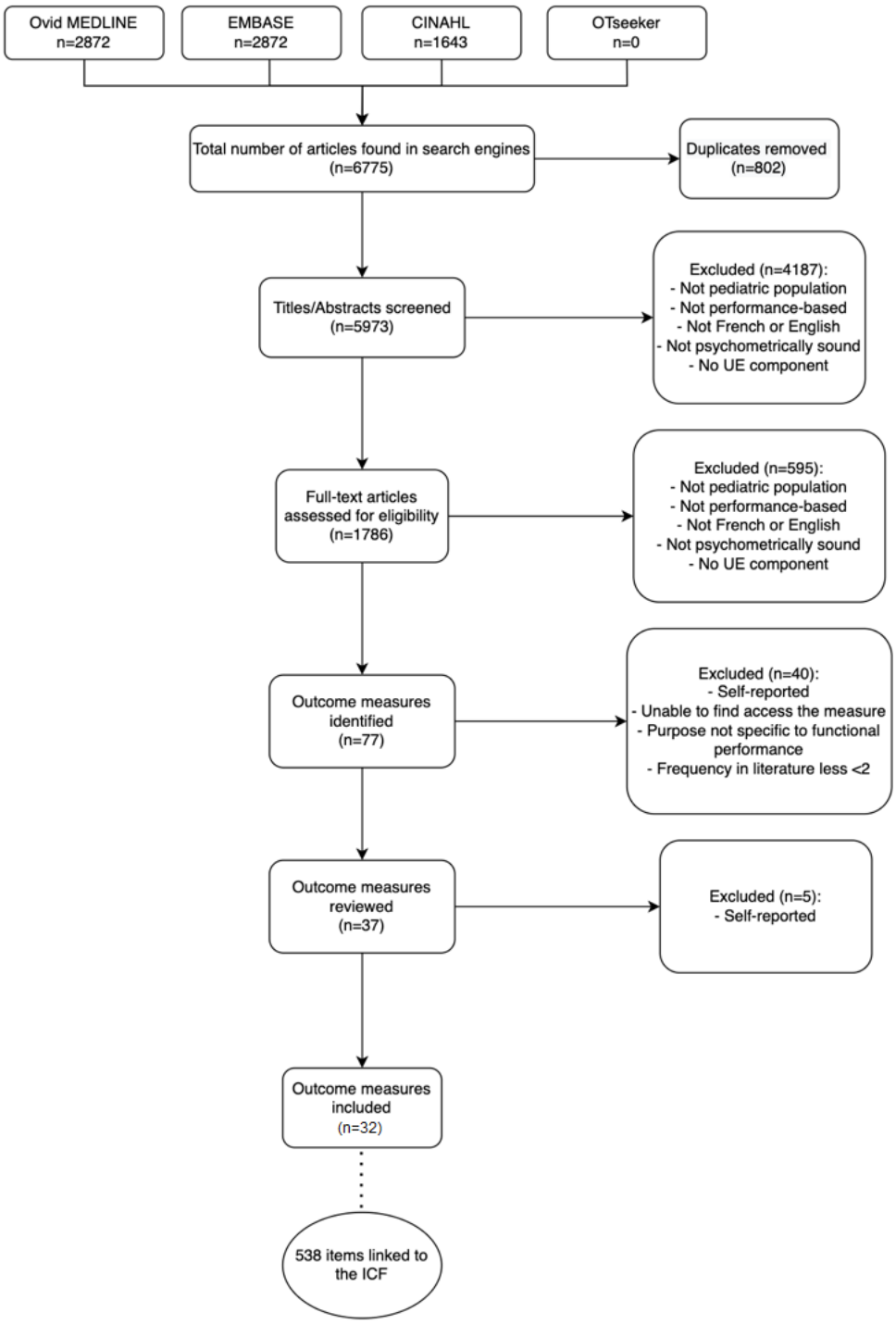


Figure 2. Pie chart for Assisting Hand Assessment (AHA) codes

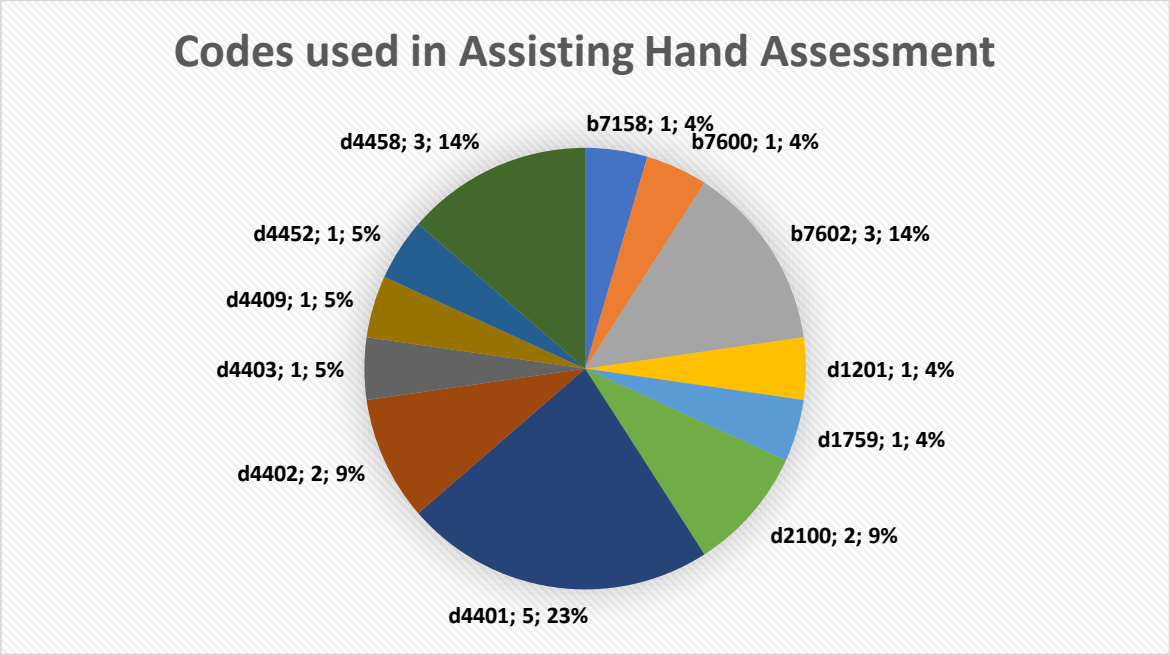
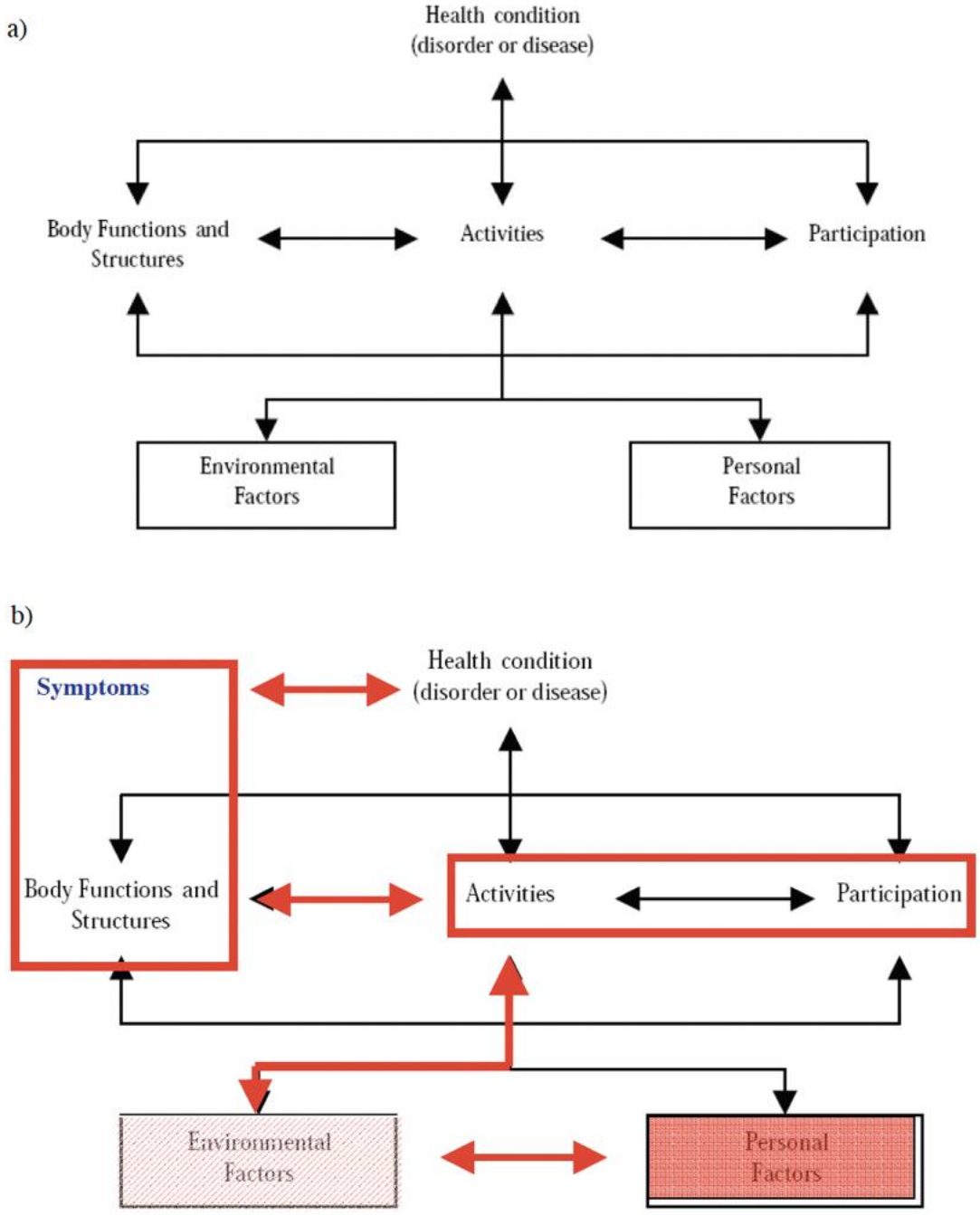


Figure 3. Differences between ICF as a model of functioning and as a coding system (Salvador-Carulla & Garcia-Gutierrez, 2011)



Theoretical interaction of the components of the "process of functioning" model of the ICF b) actual relations among the different components in the ICF classification system (environmental factors are partially described, whilst personal factors are not coded by the ICF).

4.11 References

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Chapter 5: Manuscript 2

5.1 Integration of Manuscripts 1 and 2

Prior to conceptualizing this thesis, a preliminary study was conducted to understand the multifaceted needs of youth with AMC, their caregivers, and clinicians [68]. The results revealed that clinicians voiced a need for the development of a condition-specific outcome measure and rehabilitation practice recommendations. In addition, during the 2nd International Symposium on Arthrogryposis in 2014, a need for the development of a standardized assessment of short- and long-term outcomes was identified [79]. However, according to the youth with AMC, the need that was identified was to prioritize participation interventions [68]. In order to further explore the participation needs of youth with AMC, I conducted another scoping review. Because the literature was so limited in this field, we found most of the information on participation on YouTube videos posted online directly by families of children with AMC. The findings indicated that rehabilitation interventions should target participation and address environmental barriers [70]. Traditionally, rehabilitation may have focused primarily on physical function and symptom management. However, based on the results of the two projects identified, the ultimate goal of rehabilitation should be to enable individuals to fully participate in society and engage in meaningful activities.

Consequently, the second and third studies of this thesis aimed to engage various stakeholders such as caregivers and medical experts in the development process of the AMC-specific outcome measure. This collaborative approach ensured that the SHAPE-UP would be comprehensive, relevant, and sensitive to the unique needs of children and youth with AMC.

5.2 Manuscript Title Page

TITLE: Stakeholder Engagement in the Development of an Upper Extremity Outcome Measure for Children with Rare Musculoskeletal Conditions

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Research Involvement and Engagement

RESEARCH Open Access

Stakeholder engagement in the development of an upper extremity outcome measure for children with rare musculoskeletal conditions

Caroline Elfassy[1,2*], Lisa Wagner[3], Johanne Higgins[4], Kathleen Montpetit[2], Laurie Snider[1] and Noémi Dahan-Oliel[1,2]

Abstract

Background Upper extremity (UE) involvement is prevalent in 73% of individuals with arthrogryposis multiplex congenita (AMC), yet no AMC-specific outcome measure exists. When developing a measure specific to a population with a rare musculoskeletal condition, clinicians' and patients' perspectives and involvement is a crucial and necessary step. This study sought to determine the most clinically useful items for an outcome measure of UE function for children with AMC as defined by caregivers and clinicians.

Methods To ensure the perspectives and needs of caregivers of children with AMC and clinicians were considered in the development of the UE measure for AMC, a Nominal Group technique (NGT) with caregivers of children with AMC (phase 1) followed by a three-round survey with clinicians (phase 2) were carried out.

Results Phase 1: Eleven individuals participated in the nominal group technique and identified 32 items. The most important items were Picking up an object (n = 11), Eating (n = 10), Reaching mouth (n = 10), Getting out of bed (n = 10). Phase 2: Invitations to participate to an online survey was sent to 47 experts in the field of AMC, 20 participants completed round 1, 15 completed round 2 and 13 completed round 3. Throughout the survey, participants were asked about movement required to screen the UE, essential domains to be included in the measure, establishing a scoring guide and identifying tasks associated with joint motion and position.

Conclusion A preliminary version of an UE AMC-specific outcome measure was developed with the help of caregivers' perspectives and expert opinions.

Keywords Stakeholder engagement, Outcome measure development, Rare musculoskeletal condition, Upper extremity

5.3 Abstract

Background: Upper extremity (UE) involvement is prevalent in 73% of individuals with arthrogryposis multiplex congenita (AMC), yet no AMC-specific outcome measure exists. When developing a measure specific to a population with a rare musculoskeletal condition, clinicians' and patients' perspectives and involvement is a crucial and necessary step. This study sought to determine the most clinically useful items for an outcome measure of UE function for children with AMC as defined by caregivers and clinicians.

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Conclusion: A preliminary version of an UE AMC-specific outcome measure was developed with the help of caregivers' perspectives and expert opinions.

Plain English Summary:

Arthrogryposis multiplex congenita (AMC) is a rare musculoskeletal condition that affects the joints and muscles of the body. In about 70% of the cases, it affects the upper extremities (UE). However, there is no specific outcome measure for UE function in children with AMC. An outcome measure refers to a tool or method used to assess and measure the results or effects of a particular treatment, intervention, or condition. It helps healthcare professionals and researchers understand the impact or outcome of a specific situation, such as the level of improvement or changes in a person's health or function. The goal of this study was to develop such a measure while accounting for the perspectives of youth with AMC, their caregivers and clinicians.

To achieve this, a study was conducted in two phases. In the first phase, a Nominal Group technique (NGT) was used to gather input from caregivers of children with AMC. Eleven individuals participated and identified 32 items, with the most important being picking up an object, eating, reaching the mouth, and getting out of bed.

In the second phase, a three-round survey was sent to 47 experts in the field of AMC, with 20 participants completing the first round, 15 completing the second round, and 13 completing the third round. The survey asked participants about screening UE movement, essential domains to include in the measure, establishing a scoring guide, and identifying tasks associated with joint motion and position.

With the help of caregivers' perspectives and expert opinions, a preliminary version of an UE AMC-specific outcome measure was developed. This measure will be useful in assessing the UE function in children with AMC and will aid clinicians in developing appropriate treatment plans for this rare condition.

5.4 Introduction

In the last 10 years, patient engagement in health research has emerged as the next evolution in healthcare delivery (1–3). The Canadian Institute for Health Research's Strategy for Patient Outcome Research (SPOR) defines patient engagement in health research as "occur[ing] when patients meaningfully and actively collaborate in the governance, priority setting, and conduct of research, as well as in summarizing, distributing, sharing, and applying its resulting knowledge" (4). Involvement of key stakeholders, which includes patients, in the development of outcome measures to be used in clinical research is essential (5,6). Indeed, de Vet et al. (2011) state that instruments to measure functioning should be developed in close cooperation with experts (i.e. clinicians who have extensive expertise on target population or patients' lived experience) (7). When developing a measure specific to a population with a rare musculoskeletal (MSK) condition, clinicians', patients' and caregivers' perspectives and involvement is a crucial and necessary step. One such MSK condition is arthrogryposis multiplex congenita (AMC).

AMC is a term used to describe a group of congenital conditions characterized by joint contractures in two or more body areas (8). Treatment involves rehabilitation and orthopedic surgery with multidisciplinary team members to maximize the capacity and performance of the child with AMC to provide the best outcomes throughout the lifespan (9). The multidisciplinary

team consists of the child and their family with varied healthcare professionals (e.g., orthopedics, genetics, neurology, rehabilitation) depending on how severely the child is affected. AMC can affect the upper and/or lower extremities as well as other body areas such as the gastrointestinal, genitourinary and central nervous systems (10). The lower extremities are involved in 89-95% of cases (11), while individuals have upper extremity (UE) involvement in about 73% of cases (12). The most frequent clinical presentation of the UE at birth has been described as internal rotation of the shoulders, extension of the elbows, flexion of the wrists, thumb-in-palm deformity, and variable movement in the fingers (12,13). According to Hamdy et al. (2019), UE function is the most determining aspect of quality of life and independent living for individuals living with AMC as it pertains to daily activities such as dressing, perineal hygiene, grasping, use of mobility aids when needed, and feeding (11).

According to Wagner et al. (2019), rehabilitation practitioners (i.e., occupational therapists (OTs) and physical therapists (PTs)) enable function and help individuals with AMC participate in meaningful activities (14–16). Outcome measures can be defined as a tool or method used to assess and measure the results or effects of a particular treatment, intervention, or condition. The use of outcome measures can help clinicians evaluate a child's capacity and performance and determine a treatment plan based on the child's needs. OTs and PTs use a variety of outcome measurement and evaluation tools to assess a patient's level of functioning. Although many outcome measures exist, some were developed specifically to assess and evaluate a region of the body and others were tailored for specific pediatric populations (17).

Currently, clinicians utilize generic measures aimed at overall assessment of functional performance and/or motor function as there are no AMC-specific outcome measures that exist. These standardized measures provide important information on the child's level of function. However, they were not developed for children with an UE deformity, do not inform as to which specific joint or muscle limits functional performance, and may not reflect the adaptations or compensatory strategies used by children with AMC (for example, using feet or mouth for activities). For example, the Functional Dexterity Test was developed to measure manual dexterity skills for functional fine motor coordination tasks performance in adults and children (18) whereas the Shriners Hospital Upper Extremity Evaluation (SHUEE) was designed to evaluate UE function in hemiplegic cerebral palsy in children between 3 and 18 years of age. Although some generic UE outcome measures have been used with the AMC population, these have not been validated

for a standardized evaluation (19). Indeed, during the Second International Symposium on Arthrogryposis held in St. Petersburg, Russia in 2014, a need for the development of a standardized assessment of short- and long-term outcomes was identified (20). The authors explored what is currently known regarding participation among children and youth with AMC (21) as well as identified the needs surrounding rehabilitation according to youth with AMC and caregivers (22). In a preliminary study on the development of a standardized AMC-specific outcome measure, the authors identified an item bank of the most frequently reported pediatric performance-based outcome measure (PBOM) of UE function and linking their content to the International Classification of Functioning, Disability and Health (ICF) (Elfassy et al. (Submitted). In the scoping review, the authors defined UE function as UE coordination, motor function, sensation, muscle strength, and stabilization. In order to engage caregivers and clinicians, the aim of this study consisted of identifying, according to their expertise, the most clinically useful items for an outcome measure of UE function for children with AMC. The ICF framework and definitions were used exclusively for all phases of the development of the UE AMC-specific outcome measure.

5.5 Methodology

This study received institutional approval in May 2021 (CAN2103) and ethics approval from the institutional review board of McGill University's Faculty of Medicine in May 2021 (A03-B15-21A)

To ensure the perspectives and needs of caregivers of children with AMC as well as clinicians were considered in the development of the UE measure for AMC, a Nominal Group technique (NGT) with caregivers of children with AMC (phase 1) followed by a three-round survey with clinicians (phase 2) were carried out. These two phases are described below.

Phase 1: Patient Engagement Nominal Group Technique

The NGT is a structured face-to-face small group discussion aimed at reaching consensus and providing a prompt result for researchers (23,24). The NGT gathers information by asking individuals to respond to questions posed by a moderators, and then asking participants to prioritize the ideas or suggestions of all group members (23). The four steps used for the NGT included : 1) generating items, 2) recording items, 3) discussing items, and 4) voting on items (23–25). During a breakout session at the 14th Annual Arthrogryposis Multiplex Congenita Supper Inc. (AMCSI) Conference in July 2019 in Norfolk, USA, youth and adults with AMC and their caregivers were invited to participate

in an open group discussion to gather and exchange ideas regarding the future development of an UE outcome measure specific for children with AMC. As AMC is comprised of a group of heterogeneous conditions with varying levels of severity and involvement, any individual presenting with multiple congenital contractures as well as other comorbidities such as CNS involvement and their caregivers were invited to participate. The session was audio-recorded, and interested participants were asked to verbally consent to participate.

The meeting took place in a closed room where all participants were able to talk freely and confidentially. An opening statement where an agenda as well as the importance of each member's contribution was presented by PowerPoint. In order to address the first step of the NGT, the overall statement presented looked to answer the following:

> "When a therapist or rehabilitation professional is assessing you or your child's arm/shoulder/elbow/wrist/hand/finger, what do you think is an important aspect or item to consider in this evaluation (it could be a simple task or an activity that requires many steps."

Each participant was asked to answer the above statement, with as many responses, silently and independently, on a piece of paper provided by the research team. The second step consisted of engaging all participants, one at a time, in a round-robin feedback session to concisely record each item (without debate). The round-robin continued until each participants' items had been documented. The third step entailed discussing each item to determine the clarity and importance. For each item, the principal author asked participants if they had any questions or comments regarding the item or if they required clarification. Finally, the fourth step involved voting on the items generated. The voting consisted of asking each participant to categorize each of the identified items as A) Important and essential, B) Important but not essential, and C) not relevant. Each participant voted independently.

Phase 2: Clinician Opinion using a Three-round Survey

The survey consisted of pre-selected items drawn from preliminary work (21,22,26) as well as the findings of the NGT. The participants included in the survey were clinicians as phase 2 focused not only on the development of items but also on creating a scoring guide to be used during clinical evaluations. Approximately 50 clinicians in the field of AMC were invited to participate in a three-round survey. These clinicians included PTs, OTs, certified hand therapists, orthopedic surgeons, and physical rehabilitation technicians. The clinicians were identified during the 3rd

International Arthrogryposis Symposium in Philadelphia, USA, in 2018. Invitations to participate in the survey process were sent by e-mail, explaining the purpose of the project, with a link to an electronic survey using the Qualtrics online platform. Participants were asked to complete the survey within a 1-month timeframe. A weekly reminder was sent using the Qualtrics software. Clinicians were eligible to participate regardless of location or setting of practice, as long as they had at least 2 years' experience working with the pediatric AMC population. A summary of the surveys and can be found in Appendices 3,4, and 5.

Based on findings of preliminary work (21,22,26), the authors prepared a draft version of the survey which included the purpose of the measure, the inclusion/exclusion criteria, consent waiver link, demographic information, as well as "must have" items of the outcome measure to be included.

The first-round of the survey asked each clinician a series of demographic questions (i.e., profession, country of residence, type of work setting, years of practice, years of experience with AMC, number of children with AMC they have worked with, healthcare professionals on their team). In addition, clinicians were presented a series of questions related to domains of performance-based items and domains of scoring such as range of motion. Similarly to Lawshe's method of content validation, expert clinicians were asked to rate each item as essential, useful but not essential, and not necessary (27). A comment box was made available throughout the survey and for each item should the participants have additional comments to raise. Invitations to participate in the second and third round survey were only sent to clinicians who had previously completed or partially completed the prior rounds.

During the second-round survey, clinicians were presented with the preliminary version of the purpose of the measure and the scoring guide. Clinicians were asked to determine item clarity and word choice for the different subtasks. They were also asked which joints should be the focus as well as the hierarchy of scoring used for analysis of joint motion and position score.

For the third and final round, the clinicians were presented with the final version of the purpose of the measure and the scoring guide. Clinicians were asked to look at the measure as a whole and provide any comments regarding scoring, overall layout and clarity of content.
Descriptive analysis was provided for both phases, particularly the three-round surveys. Results arising from partially completed surveys were not included and were considered as non-complete for our analysis.

5.6 Results

Phase 1: Patient Engagement Nominal Group Technique

During the 14th Annual Arthrogryposis Multiplex Congenita Support Inc. (AMCSI) Conference, a total of 11 individuals (7 mothers, 3 fathers, and 1 adult with AMC) participated in the NGT and identified 32 items. The detailed list of identified items can be found in Table 1. According to the voting system, the most important and essential items identified were the following: Picking up an object (n=11), Eating (n=10), Reaching mouth (n=10), Getting out of bed (n=10), Using spoon/fork (n=9), Putting on pants (n=9), Dressing (donning and doffing coat) (n=9), Brushing teeth (n=9), and Ability to participate in social games with family members (n=9).

Phase 2: Clinician Opinion using a Three-round Survey

Of the 47 clinicians invited to the first-round of survey, 20 completed the first round, 15 the second round, and 13 completed the third round. Demographic information of the participants for each completed survey round can be found in Table 2.

The first-round survey questionnaire can be found in Appendix 3. The clinicians (n=20) had on average 17.86 (2-35 years) years of experience working with children and youth with AMC. They oftentimes worked with a multidisciplinary team consisting of pediatricians (n=5), orthopedic surgeons specializing in different limbs (n=19), nurses (n=15), orthotists (n=17), OTs (n=17), PTs (n=20), physiatrists (n=3), geneticists (n=10), neurologists (n=6), social workers (n=13), psychologists (n=5), speech language pathologists (n=4), and neurosurgeons (n=2). The most important movements for UE screening according to the clinicians were elbow flexion, forearm supination, wrist extension, and finger flexion. Other important UE screening movements can be found in Table 3. Additionally, based on the results of the first-round survey, the domains deemed to be essential to be evaluated in the UE outcome measured included UE weight bearing, UE to change body position, grasp, release, reach, fine hand manipulation, dressing, feeding, and toileting. The Throwing and Catching domains were deemed useful but not essential by the clinicians. Specific items can be found in Table 4.

Based on the results from the first round, the preliminary version of the UE outcome measure was created. In this second survey round (Appendix 4), the purpose of the measure was revised (i.e. describe impairments, activity limitations, and participation restrictions in the performance of daily tasks in children with AMC to guide treatment decision-making and

evaluation of treatment effectiveness for the UE), a scoring guide for task completion was developed (Table 5), 12 tasks and 3 descriptive questions (Table 6) were generated, and a scoring table including analysis of joint motion and position was derived. Fifteen clinicians completed the survey, three partially completed it, and two did not respond. Clinicians in the second-round survey were asked about terminology and which joints to analyse for each task. Table 7 presents the different terminology choices offered to the clinicians. Based on the scoring provided by the clinicians, additional joints such as wrist in task 2, forearm in task 4, thumb, fingers, and wrist in task 6 and 7, fingers, wrist, forearm, and shoulder in task 10 and shoulder in task 12 were added to be analyzed in the scoring sheet.

The third and final round survey was sent to 18 participants (Appendix 5), 13 participants completed the survey in its entirety and 5 partially completed it. Modifications to the UE outcome measure included substituting the arc of motion of a joint to a specific direction of movement (i.e. no external rotation, partial external rotation, full external rotation vs. internal rotation, neutral, external rotation), modifying the hierarchy of scoring, and adding a scoring row to reflect a specific item (i.e. reveal the bimanual nature of the task (e.g. stabilizing with one hand and fold/cut with another). Task 12 was removed as it was replaced with a table consisting of various transfers capacities that the child can perform (i.e. bed positioning, lying to sitting, sitting to standing, toilet transfer, bathtub/shower transfer, getting in and out of a car) in the descriptive questions section (Table 8). Task 12 was no longer specific to only transferring from lying to sitting but rather assessing all transfers required in daily routines. The qualitative questions were edited to include propelling a manual wheelchair and operating a motorized wheelchair.

5.7 Discussion

The goal of this study was to describe the process of developing a new outcome measure of UE function for children with AMC and identify the most clinically useful items to be included as determined by individuals with AMC and their caregivers, and clinicians. The results from the NGT and the survey yielded a first of its kind preliminary version of an AMC-specific UE outcome measure. The outcome measure includes 11 tasks with a total of 47 subtasks and 3 qualitative descriptive questions. A complete scoring guide as well as an analysis of joint motion and position was developed.

According to the findings in phases 1 and 2, the items identified were predominantly related to the Body Functions and Structures and the Activity domains of the ICF. This result is not entirely surprising as specific outcome measures related to other pediatric conditions have shown the same results. For example, in a previously published systematic review, the authors aimed to define and link the meaningful concepts of items contained in three commonly administered standardised UE outcome measures (i.e. Melbourne Assessment, Quality of Upper Extremity Skills Test (QUEST), Assisting Hand Assessment (AHA)) used in cerebral palsy (28). According to the review, the Melbourne Assessment was reported as a measure of the Activity domain of the ICF, the QUEST was related to both the Body functions and Activity domains of the ICF, and the AHA was consistent with the Activity domain of the ICF (28). Additionally, an article showcasing the overview of assessments and classification tools used to understand and measure UE function associated with children with spasticity indicated that the Kids-Assisting Hand Assessment, Mini-Assisting Hand Assessment, Children's Hand-use Evaluation Questionnaire, ABILHAND-Kids, Canadian Occupational Performance Measure, and Goal Attainment Scaling were linked to the Activity domain of the ICF, the Pediatric Motor Activity Log was related to Body Function and Structure domain of the ICF, and the Melbourne Assessment, QUEST, Box and Block of Manual Dexterity, SHUEE were related to Body Function and Structure and Activity domains of the ICF (29). Pediatric specific UE outcome measures used in clinical settings across different diagnoses have been shown to be focused more on the Body Functions and Structures and Activity Domains of the ICF. Indeed, there was a need surrounding the Participation and Environment domain of the ICF that was identified in a recent publication (22). Youth with AMC stated that rehabilitation focused primarily on physical limitations which did not always correspond to the youth's specific participation needs (22). Therefore, the throwing and catching subtasks were kept in the measure even though they were deemed useful but not essential in the survey, as it was important to consider participation based on the needs identified by key stakeholders, in this case individuals with AMC.

The clinical implications of involving both individuals with lived experience as well as clinicians with expertise in the field of AMC in the development of an outcome measure is twofold. The first implication is having a complete picture of the needs surrounding the evaluation and intervention provided to the child with AMC. Phase 1 of this project (i.e. NGT) allowed for patient engagement and highlighted the importance of including the Participation domain of the ICF in the development of UE outcome measure. Involving youth and caregivers in the development of

outcomes and clinical research has been shown to be feasible and valuable to studies (30). The second clinical implication is to help increase awareness and public interest of children and youth with rare musculoskeletal conditions. Establishing a network of patient partners and clinicians in rare diseases is important to help increase the quality of studies being published and disseminating the results to the population in question.

5.8 Conclusion

A preliminary version of an UE AMC-specific pediatric outcome measure was developed with the contribution of patient engagement and clinicians' opinion on 11 task and 3 descriptive qualitative questions. The measure includes a scoring guide for task completion as well as a joint motion and analysis section to determine which joints of the UE are limiting the child's with AMC capacity to complete the task.

5.8.1 Limitations

Although youth with AMC and caregivers were invited to participate in the NGT at the 14th Annual AMCSI Conference, only the caregivers attended the breakout session. However, findings from previously published studies regarding the needs of youth with AMC (21,22) were included in the development of the preliminary version of the outcome measure.

Due to the scope of study, initial validation of the measure could not be completed at this current time. While this is the first step towards finalizing a pediatric AMC-specific UE outcome measure, further assessment of reliability, construct validity and responsiveness is required and is planned with the next phase of this project. The development of a psychometrically sound UE measure in AMC will elevate current practice and assist in establishing the effectiveness of surgical and non-surgical therapies.

5.8.2 Acknowledgement

We wish to acknowledge the youth, caregivers, and clinicians who contributed in the preliminary phases of this project as well as those who participated in the stakeholder engagement portion of the project.

5.8.3 Declaration of Interest

The authors declare that they have no competing interests.

5.8.4 Funding

This project received seed funding by “Partenariat de recherche clinique en ergothérapie OEQ-REPAR (2017-2020), followed by a multisite grant from Shriners Hospitals for Children (2021-2023). C.E is supported by a “Fond de recherche du Quebec – Santé” Doctoral Training for Applicants with a Professional Degree Award. N.D-O is supported by a “Fond de recherche du Quebec – Santé” Clinical Research Scholar Junior 1 Award.

5.8.5 Data Availability

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

5.8.6 Ethical Considerations

This study received institutional approval at the Shriners Hospitals for Children – Canada in May 2021 (CAN2103) and ethics approval from the institutional review board of McGill University’s Faculty of Medicine in May 2021 (A03-B15-21A). Consent to participate was received for phase 1 (patient engagement) and 2 (clinician opinion).

5.9 Tables

Table 1. Items pool during the Patient Engagement Nominal Group Technique

Item #	Item Name
1	Ability to participate in social games with family members (board games, ball catch)
2	Ability to participate in recess at school with peers (elementary)
3	Ability to use adapted objects
4	Autonomy (individualized goals)
5	Putting on pants
6	Satisfaction with completion of tasks
7	Eating (holding a bottle)
8	Using spoon/fork
9	Range of motion of shoulder, elbow, wrist, and fingers
10	Constantly re-evaluating
11	Everyday living
12	Milestones-appropriate goals

13	Holistic, global approach
14	Picking up an object
15	Writing
16	Bilateral hand use
17	Active vs. passive ROM
18	Reaching mouth
19	Playing at midline
20	Throwing
21	Dressing (donning and doffing coat)
22	Brushing teeth
23	Getting out of bed
24	Natural vs. clinical environment
25	Completing activities per age (developmental milestones)
26	Routine breakdown
27	Preparing food
28	Driving a car
29	Opening a door using a key
30	Managing hair
31	Light switches
32	Scoring and recommendations

Table 2. Demographic information of the experts the Three-Round Survey.

Round 1 (n=20)			Round 2 (n=15)			Round 3 (n=13)		
Occupation	Country	Work Setting	Occupation	Country	Work Setting	Occupation	Country	Work Setting
Orthopedic surgeon (n=9)	USA (n=13)	Hospital (n=16)	Orthopedic Surgeon (n=6)	USA (n=10)	Hospital (n=12)	Orthopedic surgeon (n=4)	USA (n=8)	Hospital (n=11)
PT (n=4)	Canada (n=3)	University (n=1)	PT (n=2)	Canada (n=2)	Speciality Hospital (n=2)	PT (n=4)	Canada (n=2)	University (n=1)
OT (n=6)	Poland (n=2)	Speciality Hospital (n=2)	OT (n=7)	Poland (n=1)	Rare Disorder Center (n=1)	OT (n=5)	Poland (n=1)	Rare Disorder Center (n=1)
CHT (n=1)	Sweden (n=1)	Rare Disorder Center (n=1)		Sweden (n=1)			Sweden (n=1)	
	Norway (n=1)			Norway (n=1)			Norway (n=1)	

OT = occupational therapist, PT = physical therapist, CHT = certified hand therapist

Table 3. Most important movement for the upper extremity screening rated by 20 experts (Round 1 Survey).

Shoulder	Elbow	Forearm	Wrist	Fingers	Thumb
Flexion (n=19)	Flexion (n=20)	Supination (n=20)	Extension (n=20)	Flexion (n=20)	Opposition (n=18)
External rotation (n=19)	Extension (n=15)	Pronation (n=16)	Flexion (n=17)	Extension (n=17)	Extension (n=16)
Abduction (n=17)					Flexion (n=15)

Table 4. Results of essential domains and items rated by 20 experts (Round 1 Survey).

Domains	**Essential Items**
UE weight bearing	uses non-dominant hand to stabilize self (n=10)
	uses upper extremity to weight bear in prone (n=9)
	uses upper extremity to weight bear in sitting with hands forward (n=9)
using the UE to change body position	pushes self from supine to sitting (n=18)
	using UE to use mobility aid (n=16)
	pushes self from sitting to standing (n=14)
	transferring from one body position to another (n=14)
Grasp	picking up a medium object (shoes, water bottle, plate etc) (n=20)
	picking up a small object (coin, bead, pencil, etc) (n=17)
Release	releasing a medium object (shoes, water bottle, plate, etc) (n=17)
Reach	reaching for an item from waist level (n=16)
	reaching for an item by crossing the midline (n=12)
Fine Hand Manipulation	write your name or draw something (n=20)
	opening a jar (n=15)
	closing a jar (n=12)
Dressing	puts on clothes over-head (shirt, sweater, hat) (n=20)
	pulls on pants (n=20)
	pulls down (remove) pants (n=20)
	removes shirt (n=19)
	puts on shoes (n=17)
	puts on open shirt (n=16)
	pulls up zipper (n=15)
	starts a zipper (n-=14)

	puts on socks (n=14)
	removes shoes (n=14)
	buttoning (n=13)
	unbuttoning (n=13)
Feeding	reaches mouth (n=20)
	picks up food using fork (n=17)
Toileting	places sticker on lower back (proxy for reaching to wipe buttocks) (n=19)

Table 5. Scoring guide for task completion (Round 2 Survey)

Score	Description	Example
0	Unable	The child is unable to complete any component of the task.
1	Partial completion of task passively	The child can partially complete the task using passive range of motion.
2	Partial completion of task actively	The child can partially complete the task using active range of motion.
3	Completion of task passively	The child can complete the task using passively range of motion.
4	Completion of task actively	The child can complete the task using active range of motion.

Table 6. Descriptive questions developed in Round 2 Survey

1. Does the child use their arms for using a mobility device?
2. Does the child use their arms for shifting/changing/moving body (getting on or off couch/toilet)?
3. Are you using a splint for the tasks included?

Table 7. Task terminology choice and rating (n=15) (Round 2 Survey).

Task	Terminology choice
1. Grasp/pick up a cheerio, bring it to your mouth, place it back down in front of you, and release it/let it go.	Pick up (n=14)
	Let it go (n=9)
2. Grasp/pick up a water bottle/can, bring it to your mouth, place it back down in front of you, and release it/let it go.	Pick up (n=9)
	Water bottle (n=13)
	Let it go (n=9)
3. Open the jar, pour out a few beads/macaroni/buttons, string 3 together, and close the jar.	Beads (n=13)

4. Pick up the crayon/marker, write your name on this piece of paper, fold the paper, and cut it using the scissors.	Marker (n=12)
5. Pick up the Play-Doh using the fork and bring it to your mouth.	N/A
6. Reach for a small-size ball (e.g., tennis ball) placed on the floor, throw the ball underhand. Repeat task, throwing ball overhead.	N/A
7. Reach for a medium-sized ball (e.g., basketball) placed on the floor, throw the ball underhand. Repeat task, throwing ball overhead.	N/A
8. Put on a T-shirt overhead and take off the T-shirt.	N/A
9. Put on vest/sweater with zipper, fasten the zipper, pull it all the way up, and pull it back down.	Vest (n=9)
10. Pull down your pants, reach bum/buttocks area, place a sticker on bum/buttocks area [proxy for wiping after bowel movement], place sticker in between legs [proxy for wiping after urination], and pull pants back up.	Buttock (n=9) Buttock (n=9)
11. Put on a sock and take it off.	N/A
12. Show us how you move from lying down on your back to a sitting position	N/A

Table 8. Descriptive questions amended in Round 3 Survey.

1. Does the child use their arm to: a. Propel a wheelchair b. Use a walking aid (cane, crutch, walker) c. Other

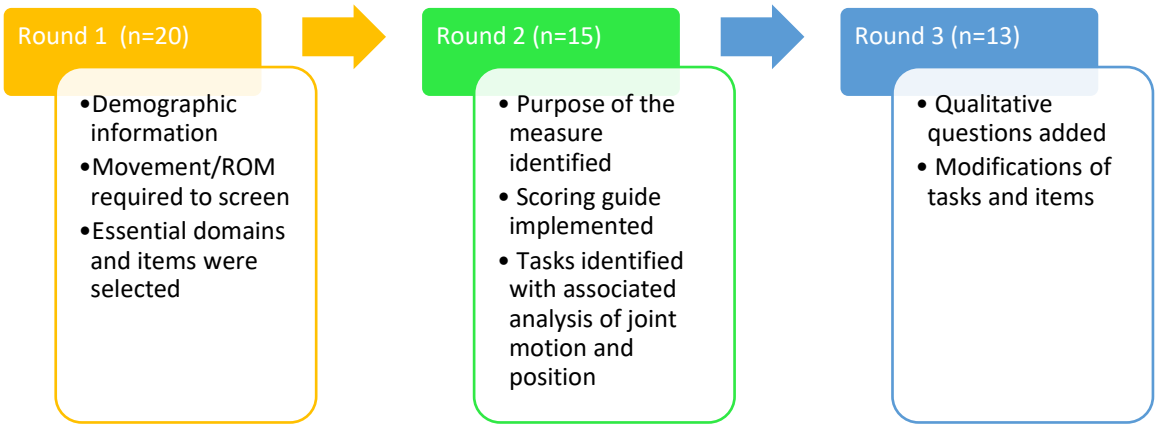
2. Does the child use their arms to perform the following transfers :

Transfers	Uses their arms		Cannot use their arms	Cannot complete due to LE contracture	Describe
	Right	Left			
Bed positioning					
Lying to sitting					
Sitting to standing					
Toilet transfer					
Bathtub/shower transfer					
Getting in and out of a car					
Other					

3. Does the child use a splint/orthosis when performing the tasks included in the SHAPE-UP?

5.10 Figures

Figure 1. Description of each survey round for the clinicians



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Chapter 6: Manuscript 3

6.1 Integration of Manuscripts 2 and 3

The initial development of the SHAPE-UP stemmed from insights gained by engaging various stakeholders, including youth with AMC, their caregivers, and experts in the field of AMC (Chapters 4 and 5). The first draft of the SHAPE-UP was developed and comprised of the Administration Manual, three descriptive questions, 11 tasks and associated subtasks, the 5-point Task Completion scale, and the Analysis of Joint Motion and Position scale. However, before clinicians were able to administer the SHAPE-UP to the AMC population, adjustments and field testing were imperative to ensure user-friendliness and the absence of potential concerns.

Over a period of ten months, bi-monthly meetings occurred among the research team to review and refine various aspects, addressing ambiguities in subtasks, the Task Completion scale definitions, and the number of joints to include in the analysis of joint motion and position. This iterative process led to the creation of five distinct versions of the SHAPE-UP. Each version was modified and further refined and the examples provided below represent some of the few modifications made during this progression.

For instance, in version 2 of the SHAPE-UP, the subtask involving "holding a string" and "beading" was added to Task 3 to illustrate the bilateral hand use required. Additionally, in version 3 some subtasks such as "Pick up a cheerio" were repeated in the scoring table to indicate a possible score for unilateral task completion (right hand OR left hand) as well bilateral task completion (right hand AND left hand) depending on how the child completed the task. In version 3, the description of the type of pinch/grasp pattern as well as any deviation (radial or ulnar) present was added to the "remarks" section found under each Task. Further enhancements were made in version 4, where additional details about the 5-point scoring of the Task Completion scale were incorporated in the scale. For example, a score of 3 was indicated where the child could complete the subtask using assistance of a body part (i.e. chest or leg), compensatory mechanism (i.e. swinging of the limb), and/or assistive device (i.e. sock aid). Refinements continued in version 5, where the 5-point scoring definitions were revised to improve clarity. Here, a score of 1 denoted "attempting to complete the task with minimal success" and a 2 indicated "actively partially complete the task with physical assistance". In Task 10, the "underhand throw" of the dodgeball was removed and replaced with "throw ball overhead" only.

Throughout this iterative process, the collaborative team's efforts ensured that the SHAPE-UP evolved into a comprehensive and effective assessment outcome measure ready to be administered to children and youth with AMC.

6.2 Manuscript Title Page

TITLE: Initial psychometric evaluation of an upper extremity Arthrogryposis Multiplex Congenita-specific measure using Rasch analysis: A cross-sectional study

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To be submitted to Archives of Physical Medicine and Rehabilitation, 2024

6.3 Abstract

Objectives: The aims of this study were 1) to evaluate the psychometric properties of the Shiners Hospital Arthrogryposis Pediatric Evaluation – Upper Extremity (SHAPE-UP) Task Completion scale using Rasch analysis and 2) determine the strength of the correlation between the number of joints involved and the Task Completion score.

Design: Cross-sectional study

Setting: Six participating Shriners Hospitals

Participants: 101 participants were recruited for the administration of the SHAPE-UP. Of these, 92 participants (54 female) completed the SHAPE-UP administration (mean=10.35 years, SD=5.18 years).

Interventions: Not applicable

Main Outcome Measure: The SHAPE-UP consists of 3 descriptive questions and seven task performance-based video-recorded evaluation. The measure was designed to focus on two scales: Task Completion and Analysis of Joint Motion and Position.

Results: The fit of the baseline data when all 46 items were considered produced a significant item-trait interaction (χ^2=129.55; p<0.05). Disordered thresholds were observed for all items except items 10 and 24. When the original five scoring options were collapsed to three options and tasks #5, 6, 8 and 9 were removed, all thresholds were ordered. This rescoring structure offered the best fit for all items and fit the Rasch model (χ^2=44.097; p=0.708). The items fit residual mean=-0.4465, Std Dev=0.7788 and the persons fit residuals=-0.656, Std Dev=1.3173. Content validity was assessed by examining the item-person map which was adequate (-10 to 7 logits). Construct validity was attained as all the items fit residuals met the requirements (all fall between ±2.5). The reliability was established as the person separation index was 0.963. There were multiple sets of items that showed high local dependency. The r was calculated to -0.0466, p-value < 0.05.

Conclusions: While the SHAPE-UP aligns with the expectations of the Rasch model, caution is advised when computing a total score for the Task Completion scale. Additional stakeholder engagement meetings are needed to carefully choose the most clinically relevant items exhibiting high correlations.

Keywords:

Pediatric, outcome measure, Rasch analysis, upper extremity, Arthrogryposis Multiplex Congenita

6.4 Introduction

Physical examination of the pediatric upper extremity (UE) includes a combination of clinical observations, formal evaluation, and patient/caregiver interview in order to provide an optimally accurate picture of pediatric UE function. When working with a pediatric population, the required movements are usually elicited through games, toys, or play [1]. Certain basic observations and movements have been standardized and developed into outcome measures. However, the UE performs an unlimited number of actions that differ from patient to patient. Thus, challenges in evaluating the UE may be due to the greater variety of tasks and activities it accomplishes [2].

According to Richards et al. (2001), an ideal measure of UE function should contain a wide spectrum of items that span the complete range of item difficulty, from the easiest tasks that can be performed by the most severely affected persons to the most difficult ones that can be accomplished only by those with near normal UE function [3]. Tasks included in outcome measures should include both unilateral and bilateral activities, consider the quality of the movement completed, and be valid and reliable to the specific population targeted [2]. Outcome measures can be divided into generic measures that are designed to be applicable to a broad range of conditions and populations, and condition-specific measures are tailored to assess specific impairments or limitations for a given population. In the context of these broad considerations for pediatric UE assessments and rehabilitation, the application of these principles becomes crucial when addressing specific pediatric conditions. Arthrogryposis Multiplex Congenita (AMC), a group of rare congenital conditions characterized by joint contractures in two or more body areas [4,5], exemplifies the need for a tailored UE outcome measure to precisely evaluate the unique challenges and functional impairments experienced by children with AMC.

Children with AMC frequently have UE involvement; approximately 56% of children have both upper and lower extremities involved, while 17% have involvement of their upper extremities alone [6,7]. Since children with AMC display variable clinical features, often affecting all joints of the UE, the development of an UE outcome measure specific to AMC would assist clinicians to better identify the joint that is most problematic and to assess the impact of the body structure-level impairment at the activity and participation level. Presently, as there exists no condition-specific measure for AMC, clinicians utilize generic measures aimed at overall assessment of functional performance and/or motor function in AMC as there exists no condition-specific

measure for AMC. However, generic measures, 1) were not developed for children with an UE deformity, 2) do not inform clinicians, youth and their families as to which specific joint or muscle is limiting functional performance, and 3) may not reflect the adaptations or compensatory strategies used by children with AMC when engaging in activities.

The development of outcome measures involves various methodologies and frameworks [8]. The use of modern psychometric methods such as Item Response Theory and Rasch can overcome limitations that arise from Classical Test Theory. Specifically, 1) the scales developed produce ordinal scores, where the difference between two adjacent scores at different points on the scale may not be equal, 2) the scores can only be interpreted for groups of patients, as measures of statistical uncertainty are only computed at the group level, and 3) the performance of scales is dependent on the particular sample in which they are used. When developing a new a scale where there is a single construct of measure, such as UE function, Rasch analysis is the most suitable.

The Shriners Hospital Arthrogryposis Pediatric Evaluation – Upper Extremity (SHAPE-UP) was developed to address gaps identified in the literature [9,10], needs voiced by youth with AMC and their caregivers [11], and expert opinion [12]. In order to validate its use in AMC, the SHAPE-UP needed to be administered, and additional testing was needed to determine if the data fit the Rasch model. The aims of this study were 1) to evaluate the psychometric properties of the SHAPE-UP Task Completion scale (i.e. content validity, construct validity, appropriateness of response categories, floor and ceiling effects, items bias, and reliability) using Rasch analysis and 2) determine the strength of the correlation between the number of joints involved and the Task Completion score.

6.5 Methodology

Ethics:

This study received institutional approval in May 2021 (CAN2103) and ethics approval from the Institutional Review Board of McGill University's Faculty of Medicine and Health Sciences in May 2021 (A03-b15-21A). Informed assent/consent was sought from patients according to site regulations, or from parents of either younger children or of youth unable to provide consent.

Outcome measure:

The SHAPE-UP, developed in 2022, was based on the World Health Organization's International Classification of Functioning, Health and Disability (ICF) Framework. Items designed to assess UE function representing the ICF domains of Body Function, Body Structures, Activity, and Participation were identified from a literature review [10] and stakeholder consultation with clinicians and people with lived experience [12]. The measure describes the results in terms of impairments, activity limitations, and participation restrictions in the performance of daily tasks in children with AMC. The SHAPE-UP consists of 3 descriptive questions and 11-task performance-based video-recorded evaluation that is administered by an occupational or physical therapist with the use of a standardized set of objects and tasks (Appendix 1 – Manuscript 3). The descriptive questions include: 1) Use of technical aids or modifications required to complete tasks of daily living 2) Use of UE to help with mobility (propelling a manual wheelchair or operating a motorized wheelchair, holding a walking aid), and 3) Use of UE to transfer (bed, lying to sitting, toilet transfer).

The videoed tasks consist of activities that elicit different skills based on the developmental stages of hand function [1,13]. During administration of the measure, the participant can complete all tasks in a dynamic fashion without starting and stopping for the administrator to record the data. The measure was designed to focus on two scales: Task Completion (i.e., can the children complete the task?) and Analysis of Joint Motion and Position (i.e., how is the child using their UE to complete the task?). The Task Completion scale is composed of 11 video-recorded tasks and includes 57 sub-tasks (46 if we remove the duplicated items that represent Right and Left UE). The Task Completion scale is originally scored from 0-5,where 0=child is unable to attempt complete or complete any part of the subtask, 1=child attempts to complete the task with minimal success, 2=child can actively partially complete the task with any physical assistance, 3=child can passively complete the task when using external aid (not physical aid from a person), compensatory movements, or a device, 4=child can actively complete the task without any external aid or compensatory movement. The Analysis of Joint Motion and Position scale contains six distinct joints of the UE (i.e., thumb, fingers, wrist, forearm, elbow, shoulder). For each of these joints, there are three specific positions to consider when scoring. To facilitate the scoring process for clinicians, only the joints targeting specific components of the tasks will be scored. This visual cue supports a more efficient and accurate evaluation. Refer to Appendices 10 and 11 for the SHAPE-UP Administration Manual and Scoring Table that was used with participants.

Procedure:

The study employed a cross-sectional design. Data were collected across North America in six Shriners Hospitals Canada, Chicago, Greenville, Northern California, Philadelphia, and Portland where the SHAPE-UP was used as a measure of UE function. Sample size, particularly when recruiting in the area of rehabilitation and rare diseases, can be a challenge. Demographic and clinical information on the included participants was collected using a case report form, and included age, sex (biologic), type of AMC, co-morbidities, description of upper and lower extremity contractures including braces worn, description of UE surgeries, level of mobility and level of independence in self-care. Children between 1 and 21 years of age who received services at one of the six participating sites and who had a clinical diagnosis of AMC described as having joint contractures to 2 or more body parts [4] and who had UE involvement were invited to participate in the study. The clinical research coordinator at each site invited eligible children and their families either during their hospital visit or remotely via telephone, recruitment flyer, or using a secured teleconferencing platform. Those who met the inclusion criteria, who were able to follow simple instructions to perform the tasks of the SHAPE-UP in French, English or Spanish and provided consent to participate were included in this study. The participating site's occupational therapist and/or physical therapist recorded the administration of the SHAPE-UP and uploaded the video to the Box system, a secure cloud content management platform. Two reviewers (C.E and L.W) watched and scored the videos of children performing the SHAPE-UP tasks accordingly using the SHAPE-UP scoring table.

Sample size:

When using Rasch analysis, sample size requirements varies depending on the item calibration, targeting and confidence [14]. Large samples are generally considered necessary for Rasch models to obtain robust item parameter estimates. However, small sample Rasch analysis is suggested as preliminary assessment of items' psychometric properties [15]. Specifically, a sample size of 100 participants would yield stable person and item estimates (±½ logit at 95% confidence) [14]. Therefore, a sample of 120 participants (about 20 participants per site) would ensure that item calibrations and person measure is stable within ± ½ logit and 95% confidence.

Analysis:

First, a principal component analysis (PCA) was performed to determine the factor structure and the dimensionality of the SHAPE-UP using SPSS Statistics (Version 29.0.1.1). Rasch

analyses were then conducted to assess how well the data fit the Rasch rating scale model. Content validity, construct validity, as well as other psychometric properties, such as the appropriateness of response categories, floor and ceiling effects, item bias, and reliability were evaluated. The RUMM2030 computer software program was used for Rasch analyses [16]. Finally, a correlation coefficient was calculated to determine the relationship between the overall Task Completion score and joint involvement.

Principal Component Analysis

The raw data obtained from the administration of the SHAPE-UP was analyzed to perform a preliminary examination of its dimensionality and factor structuring using PCA [17].

Rasch Analyses

Baseline data of the Task Completion scale of the SHAPE-UP were analyzed to evaluate their fit to the Rasch rating scale model. The Rasch rating scale model, also called the polytomous Rasch model, was developed by Andrich (1978) for polytomous data (data with $\geq$ 2 ordinal categories), when items have the same number of thresholds, and in turn, the difference between any given threshold location and the mean of the threshold locations is equal or uniform across items [16,18]. Indeed, the Task Completion scale of the SHAPE-UP is scored on an ordinal scale (originally 0 to 5). The Rasch model is used to specify an observed rating of a person on a variable of interest as a function of the ability of the person and the difficulty of the items used to derive the rating, where both are defined by their location on a continuum from least (easiest) to most (hardest) [19]. The RUMM2030 software provides an "item-person map" to showcase this relationship where the pink blocks represent groups of patients and their ability level while the blue blocks represent the item locations and their distribution. Easy items are located towards the left of the graph while difficult items are at the right. Likewise, persons who exhibit lower UE function ability are located towards the left and those exhibiting higher levels are located towards the right (see Figure 3).

Ideally, for items to "fit" the model and for the SHAPE-UP to have adequate content validity, items should be spread evenly on the continuum of difficulty level and have a wide range (from at least -3 to +3 logits).

Construct validity is attained when persons and items meet the fit statistics requirement. This is achieved when the item and person standardized fit residuals fall between ±2.5 with a mean of 0, non-significant χ^2 (acceptable probability level > 0.05, Bonferroni adjusted) and F-statistics

(acceptable probability level >0.05, Bonferroni adjusted) [2]. Another method to verify construct validity is when none of the items display differential item functioning (DIF) or item bias [20]. Items with DIF demonstrate different probabilities depending on the group of persons being assessed (e.g. male vs. female, age group, type of AMC) and violate the property of invariance inherent to the Rasch model. This means that for the same level of risk, the scores on the items should not differ based upon differing groups [21]. Age was an important factor to consider as goals and objectives to treatment differ and evolve based upon developmental stages (i.e., infant, toddler, school-age, and teenage/adult) [9]. Threshold ordering is respected when all response categories demonstrate the highest probability of being endorsed at different levels of difficulty. Thresholds are points along a theoretical continuum of item difficulty where the probability of a person scoring either 0-or-1, and 1-or-2, respectfully, is equally likely [21]. Within each item, participants with less ability, should endorse the lower scoring category and people with more ability should endorse a higher scoring category. When the opposite happens, disordered thresholds are observed [2]. Disordered thresholds mean that the item response categories are not operating in the way it was intended.

In RUMM2030, the reliability index, also called the person separation index (PSI) indicates how well the items can discriminate persons in different levels of ability with an estimate >0.8 deemed satisfactory [22].

To assess the unidimensionality of the Task Completion scale of the SHAPE-UP, the procedure of combined principal component analysis of residuals and paired t-test was used according to Smith's method [23].

If parts of the data do not fit the model, a decision to modify (e.g., rescoring items by collapsing response categories) or reject part of the data (e.g. deleting misfitting items) needs to be undertaken [21]. Misfitting items are only considered after both local item dependency and differential item functioning (DIF) have been analyzed and no strategy can find a solution [21].

Pearson's correlation coefficient Analysis

Pearson's correlation coefficient (r) was used to determine the correlations between the number of joints involved and the maximum score obtained of the Task Completion scale. A p-value < 0.05 was considered statistically significant. A positive correlation (>0) signifies that both variables tend to move in the same direction. The closer the value is to +1, the stronger the linear relationship. A negative correlation occurs when the correlation coefficient is less than 0. Any

reading between 0 and -1 means that the two variables move in opposite directions. A coefficient of 0 indicates no correlation between the two variables [24].

6.6 Results

Participant characteristics and demographic information:

There were 101 participants that were recruited for the administration of the SHAPE-UP over a period of nine months. Of these, 92 participants (54 females (58.7%), and 38 males (41.3%)) completed the SHAPE-UP administration (mean=10.35 years, SD=5.18 years). The characteristics of the participants are shown in Table 1. The general demographic information were as follows: 7 (7.6%) children between the ages of 1 and 2 years, 13 (14.1%) children between the ages of 3 and 5 years, 27 (29.3%) children between 6 and 10 years, and 45 (48.9%) youth aged 11 and over. Type of AMC as reported by the primary caregiver using Bamshad et al's classification [25] included Amyoplasia in 48 (52.2%) children, distal arthrogryposis in 26 (28.3%) children, 4 (4.3%) with syndromic or CNS involvement, and the specific type of AMC was unknown in 14 (15.2%) children.

Principal Component Analysis

The data's ordinal nature and the presence of missing values prevented definite conclusions about the factor structure revealed by PCA. However, the analysis provided helpful insight in understanding the various domains within the construct of UE. The PCA indicated the presence of two main factors that explained 37% and 16% of the variance respectively. Despite these findings, all items were retained for the Rasch analyses of the SHAPE-UP.

Rasch Analyses

The fit of the baseline data when all 46 items were considered produced a significant item-trait interaction (χ^2=129.55; p<0.05). This is an indication that the data did not fit the Rasch model. Disordered thresholds were observed for all items except items 10 "Open Jar" and 24 "Push Arms Through". To determine whether transforming the SHAPE-UP by re-categorizing the scoring options of the Task Completion scale would improve item fit and overall fit, items were re-scored. When the original five (0-4) scoring options were collapsed to three (0-2) options, all thresholds were ordered (See difference between Figure 1 and Figure 2). The following table indicates how the different levels were merged to create the new 3-point Task completion scoring.

Task Completion Score (5-point)	Task Completion Score (3-point)
0	0
1	0
2	1
3	1
4	2

In addition to the previously described scoring process for the Task Completion scale, collaborative meetings were conducted with the research team to discern which items held the utmost clinical relevance. To ensure that the SHAPE-UP served as a valuable outcome measure for clinicians, careful consideration was given to the types of surgeries commonly performed on the UE of children with AMC, such as humeral rotation osteotomy, elbow release, tendon transfers, forearm rotation for alignment improvement, carpal wedge osteotomy, thumb-in-palm reorientation [6]. Consequently, the initial set of 11 tasks was refined to 7 tasks (removed Tasks #5, 6, 8, 9) with redundant items eliminated due to clinical reasoning (statistically all items fit the model) (e.g. bringing cheerio to mouth vs fork to mouth), and emphasis placed on tasks that highlight shoulder movement (e.g. fastening a zipper on a vest and throwing a ball overhead).

Furthermore, the Analysis of Joint Motion and Position underwent further refinement to encompass parameters such as prehension, joint limitations of the wrist, forearm, elbow, and shoulder, utilization of adaptive equipment or physical assistance, and identification of compensatory movements. This expanded scope enables clinicians to obtain more comprehensive and descriptive insights into how children utilize their UE to perform specific tasks.

After collapsing the 5-point Task Completion scale to a 3-point Task Completion scale and reducing the items from 46 to 25 items, all items fit the Rasch model (χ^2=44.097; p=0.708). The items fit residual mean=-0.4465, Std Dev=0.7788 and the persons fit residuals=-0.656, Std Dev=1.3173.

Content validity was assessed by examining the person-item threshold distribution (Figure 3). The items thresholds spread from approximately -10 to 7 logits, which is adequate.

Construct validity was attained as all the items fit residuals met the requirements (all fall between ±2.5). Additionally, for the Age Group the only items displaying variance were item 13 "Pick up bead" and item 45 "Reach buttock front". Figure 3 is also used to evaluate the floor and ceiling effects. Since there were persons located to the left and the right of the outermost items at the left and right of the graph, there were floor and ceiling effects present. Item mapping using Rasch analysis based on locations can also be found in Figure 4

The person separation index was 0.963 which indicates that the transformed SHAPE-UP can reasonably discriminate persons into different ability levels.

According to Smith's method [23], 13.33% of the t-tests are significant at the 5% level, which indicates multidimensionality.

Statistical Analysis for Analysis of Joint Motion and Position Scale:

The maximum score for the collapse 3 category Task Completion scale was 50. This indicates that a child who performed and completed all 7 tasks independently would get high score of 50. Based on the number of joints (thumb, hand, wrist, elbow, shoulder) involved bilaterally in the UE, the Pearson correlation coefficient was calculated to -0.0466, p-value < 0.05 (Figure 5). Therefore, the is a significant negative relationship between the number of joints involved and the overall score obtained in the Task Completion scale, which indicates that the more joints affected the worse Task Completion score.

6.7 Discussion

The aims of this study were to 1) evaluate the psychometric properties of the Task Completion scale of the SHAPE-UP using Rasch analysis after administering the SHAPE-UP to children and youth with AMC across six participating Shriners hospitals and 2) determine the strength of the association of the correlation between number of joint involvement and the Task Completion score of the SHAPE-UP. Rasch analysis demonstrated that the Task Completion Scale of the SHAPE-UP in its current format did not meet the criteria required for true measurement. As with many newly developed outcome measures, modifications were needed to fit the Rasch model. This included modifying the 5 point (0-4) scoring scale to a 3 point (0-2) scoring scale for the Task Completion as well as removing four tasks and certain sub-tasks. After transformation, all the items met the expectations of the Rasch model.

To our knowledge, the SHAPE-UP is the first performance-based outcome measure of UE function in children and youth with AMC that was fully developed using Rasch analysis. The SHAPE-UP was validated for content and construct validity, as well as reliability.

Although missing data lowered the validity of the fit statistics in the Rasch analysis, the estimates were not biased. However, further testing is needed to ensure the unidimensionality of the transformed version of the SHAPE-UP and to ensure its psychometric characteristics. There are signs of floor and ceiling effects as seen in Figure 3. These need to be further investigated as a floor and ceiling effect make discrimination among participants among the top and bottom end of the scale impossible. As shown in Appendix 2 – Manuscript 3, there are multiple sets of items that show high local dependency. Local independence of items is an assumption in the Rasch model which infers that the items in a test or outcome measure should not be related to each other [26]. Items should be independent of one another after considering the total. A locally dependent item is not necessarily a bad item, just a partial replication of another item, referred to as "redundancy" [20]. Even after collapsing the Task Completion Scale categories and removing tasks, there remains local dependency for a significant number of items, particularly those requiring bilateral hand skills. For example, although all items fit the model, items 19 "stabilize to cut" and 20 "cut the paper" and item 41 "Wind up to throw dodgeball" and 42 "Throw dodgeball overhead" are inherently going to be highly correlated due to the nature of the task. However, from a clinician perspective, these items were deemed useful to be included in the SHAPE-UP as the Joint Position was different between items (e.g., reaching midline at waist level vs reaching midline at shoulder level). The interdependence among these items can significantly influence the overall relationship within the SHAPE-UP and exert a considerable impact on the dimensionality of the Task Completion scale.

Regarding the DIF related with the Age Group, two potential explanations arise. Firstly, the youngest group may lack the developmental skills necessary to successfully undertake the task or subtask, rendering it "too challenging" for 1-2 year-olds. Furthermore, this age group constituted the smallest sample size with only seven participants (n=7), which is insufficient to drawing any meaningful conclusions. However, one potential solution for this challenge could involve implementing a distinct scoring guide that considers hand developmental maturity skills throughout childhood.

In the present study, a significant association was found between the number of joints involved and the Task Completion score, which indicates that the more joints are affected in the UE, the worse the score on the Task Completion scale of the SHAPE-UP.

6.8 Conclusion and Future Work

While the SHAPE-UP aligns with the expectations of the Rasch model, caution is advised when computing a total score for the Task Completion scale as certain validation estimates remain inconclusive at this point (i.e., local dependency, sample size). Additional stakeholder engagement meetings are needed to provide clinicians with the opportunity to carefully choose the most clinically relevant items exhibiting high correlations. Future steps will involve establishing the psychometric properties of the SHAPE-UP, including its inter-rater, intra-rater and test-retest reliability, convergent and discriminant validity as well as responsiveness following UE surgery. This iterative process aims to further refine the SHAPE-UP and ensure a thorough alignment with the Rasch model.

6.8.1 Limitations

This study exhibits two primary limitations. First, when recruiting individuals with rare disease, having large enough sample sizes can be a challenge. While our sample size of 92 participants is substantial for research related to AMC, it is noteworthy that it may be considered small for a thorough Rasch analysis. To confirm the psychometric properties of the Task Completion scale of the SHAPE-UP, further assessments need to be conducted on a larger and more representative sample of children and youth with AMC. Second, an important advantage to using Rasch analysis is its capacity to handle missing data effectively. However, in our study, when calculating the Pearson correlation coefficient, we were unable to address the issue of missing data in the Task Completion score.

6.8.2 Acknowledgement

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6.9 Tables

Table 1. Demographic and clinical information

Person Factor	Categories	# of Participants (n)
Age Group	1: 1 to 2 years	7
	2: 3 to 5 years	13
	3: 6 to 10 years	27
	4: 11+ over years	45
Type of AMC	1: Amyoplasia	48
	2: Distal type	26
	3:CNS involvement/syndromic	4
	4: Unknown	14
Administration Site	Canada	16
	Chicago	9
	Greenville	17
	Northern California	12
	Philadelphia	21
	Portland	17
Sex	1: Male	8
	2: Female	54
Reviewer	1: CE	47
	2: LW	45

6.10 Figures

Figure 1. Example of disordered threshold curves for item 9 “Stabilize jar” with 5 scoring options

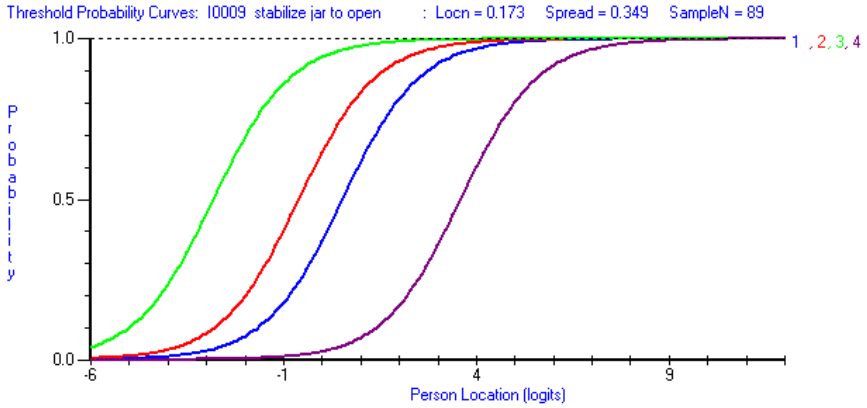


Figure 2. Example of ordered threshold ordering curves for item 9 “Stabilize jar” with 3 scoring options

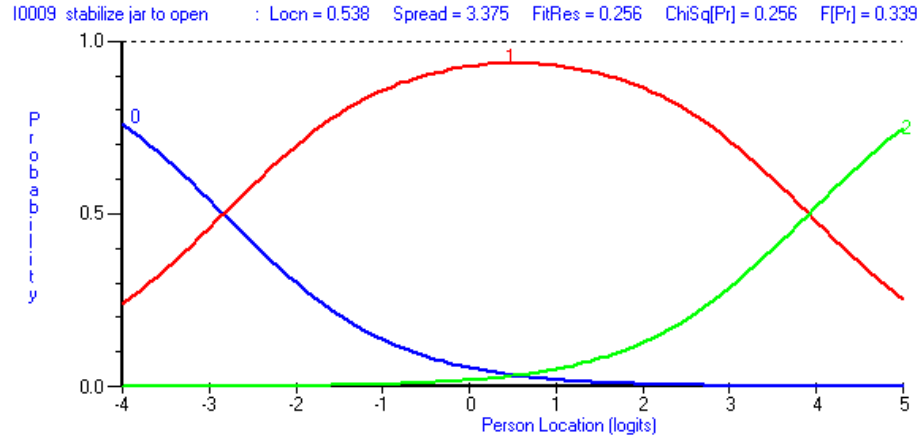


Figure 3. Person-Item Threshold Distribution

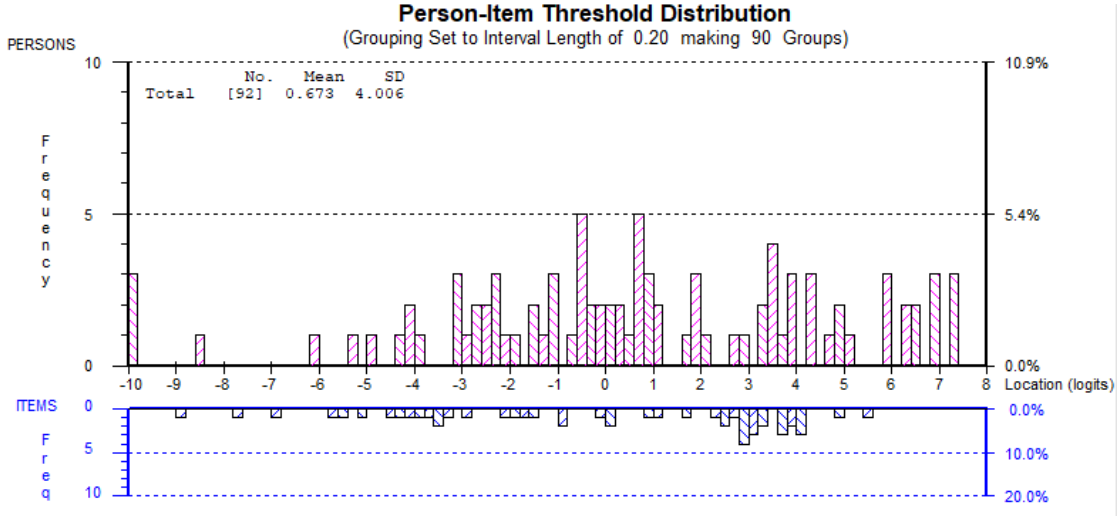


Figure 4. Item mapping using Rasch analysis

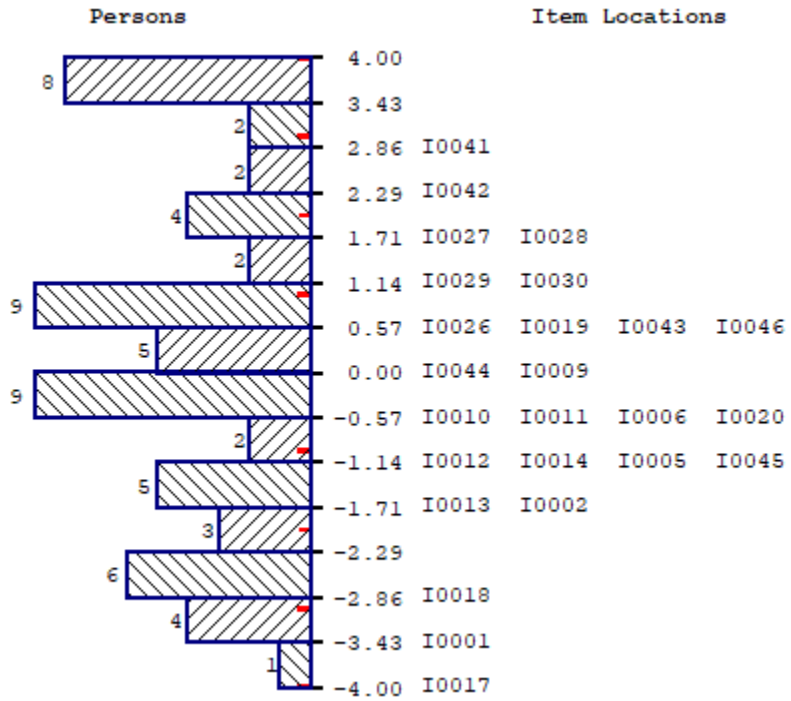
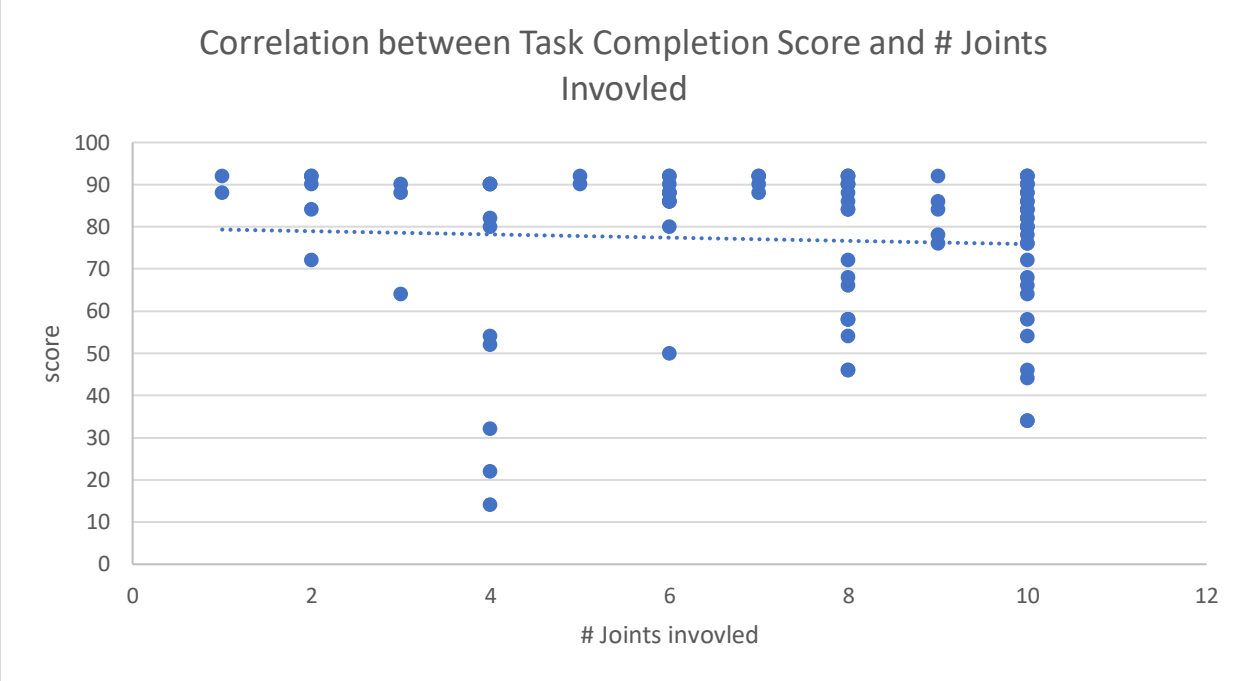


Figure 5. Correlation between Task Completion Score and number of joints involved bilaterally in the UE



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6.12 Appendices

Appendix 1 – Manuscript 3: Seven SHAPE-UP Tasks

Shriners Hospital Arthrogryposis Pediatric Evaluation – Upper Extremity (SHAPE-UP) Tasks	
1	Pick up a cheerio, bring it to your mouth, place it back down in front of you, and let it go.
2	Grasp a water bottle, bring it to your mouth, place it back down in front of you, and let it go.
3	Open the jar, pour out a few beads, pick up bead and string 3 beads onto string, and close the jar.
4	Pick up the marker, draw a circle or lines or write your name on this piece of paper with the marker, then cut or snip the paper (2-3 cuts in paper) using the scissors.
5	Put on this vest, attach the zipper, and pull the zipper all the way up and then all the way back down.
6	Throw the dodgeball overhead with both hands.
7	Pull down the shorts, reach your bottom from the back while holding 2 squares of toilet paper, also reach between your legs from the front while holding 2 squares of toilet paper, and pull the shorts back up.

Appendix 2 – Manuscript 3. Correlation matrix identifying local dependence in the Rasch model.

	I0001	I0002	I0005	I0006	I0009	I0010	I0011	I0012	I0013	I0014	I0017	I0018	I0019	I0020	I0026	I0027	I0028	I0029	I0030	I0041	I0042	I0043	I0044	I0045	I0046
I0001																									
I0002	0.181																								
I0005	0.358	0.253																							
I0006	0.135	0.233	0.477																						
I0009	-0.069	-0.037	0.017	-0.364																					
I0010	-0.17	-0.001	0.078	-0.182	0.338																				
I0011	-0.417	-0.264	-0.19	-0.259	-0.024	0.175																			
I0012	-0.265	-0.283	-0.446	-0.421	0.048	0.076	0.31																		
I0013	0.257	0.159	0.167	0.091	-0.127	-0.015	-0.272	-0.198																	
I0014	-0.184	-0.151	-0.357	-0.299	0.228	0.21	0.115	0.412	-0.157																
I0017	0.032	0.153	0.003	0.102	-0.204	-0.068	-0.124	0.25	0.244	-0.091															
I0018	0.092	0.11	0.064	0.14	-0.12	-0.07	-0.108	-0.074	0.174	-0.225	0.307														
I0019	-0.318	0.039	-0.068	-0.185	0.269	-0.078	-0.193	-0.123	-0.019	0.168	0.041	-0.093													
I0020	-0.072	0.119	-0.151	0.024	-0.044	-0.189	0.035	-0.09	-0.181	0.112	0.027	0	0.421												
I0026	-0.067	-0.034	-0.213	0.012	-0.019	-0.09	-0.05	-0.163	-0.247	-0.109	-0.146	-0.001	-0.107	-0.148											
I0027	0.127	-0.274	-0.132	-0.113	0.031	-0.155	-0.092	0.047	-0.103	-0.157	-0.318	-0.259	-0.073	-0.217	-0.008										
I0028	0.15	-0.207	-0.114	-0.128	-0.009	-0.068	-0.104	-0.005	-0.114	-0.068	-0.333	-0.276	-0.09	-0.24	0.051	0.899									
I0029	0.007	-0.327	-0.01	-0.211	-0.01	0.042	0.297	0.014	0.002	-0.001	-0.221	-0.199	-0.04	0.023	-0.147	0.37	0.367								
I0030	-0.23	-0.31	-0.291	-0.261	0.138	-0.127	0.257	0.103	-0.404	-0.079	-0.34	-0.295	0.069	0.228	0.116	0.208	0.055	0.371							
I0041	0.009	-0.243	-0.063	-0.009	-0.376	-0.367	0.001	-0.03	0.04	-0.143	-0.191	0.073	-0.269	-0.213	-0.131	-0.072	-0.102	-0.102	-0.021						
I0042	-0.12	-0.148	-0.227	-0.005	-0.122	-0.153	0.099	-0.023	-0.076	0.008	-0.271	-0.122	-0.169	-0.125	0.146	-0.079	0.01	0.048	0.135	0.526					
I0043	-0.073	-0.033	0.018	-0.095	-0.24	-0.058	-0.269	-0.1	-0.063	-0.034	-0.136	-0.274	0.017	0.034	0.054	-0.292	-0.209	-0.393	-0.195	0.087	-0.129				
I0044	0.137	-0.058	-0.116	0.021	-0.219	-0.014	0.085	-0.021	-0.084	-0.103	0.064	0.147	-0.247	-0.071	-0.024	-0.153	-0.176	-0.292	-0.247	0.105	-0.38	0.173			
I0045	0.021	-0.119	-0.038	-0.018	-0.497	-0.141	0.072	-0.036	0.242	-0.123	0.218	-0.113	-0.051	0.05	-0.152	-0.352	-0.375	-0.194	-0.22	0.025	-0.227	0.436	0.352		
I0046	-0.125	-0.047	-0.078	0.192	-0.395	-0.275	-0.232	-0.176	0.07	-0.293	-0.066	-0.12	-0.213	-0.219	0.087	0.027	-0.037	-0.27	-0.141	0.132	-0.145	0.516	0.327	0.445	

Chapter 7: Discussion and Conclusion

7.1 Summary of findings

There is a paucity of literature on UE function in AMC, itself a rare, congenital, musculoskeletal condition. Until now there existed no outcome measure specific to children and youth with AMC. Currently, clinicians utilize generic outcome measures where clinical applications are limited and not always compatible for use with children and youth with AMC.

Using stakeholder engagement, the overall aim of this thesis was to develop an UE outcome measure specific to children and youth with AMC. To achieve this overall aim, four studies comprising three manuscripts were conducted.

The first study (Chapter 4 – Manuscript 1) was a scoping review of the literature aimed at identifying which performance-based outcome measures are most frequently used to evaluate UE function in pediatric rehabilitation. The second aim was to determine the link between constructs of the ICF and meaningful concepts extracted from each measure. The results indicated that both the participation and personal factors domain were misrepresented and lacking. Findings from this review reinforced the need for stakeholder engagement in the next phase of the project.

The second and third studies (Chapter 5 – Manuscript 2) aimed at determining the most clinically useful items to be included in an UE outcome measure for children with AMC. The second study's aim was achieved through a nominal group technique. Ten caregivers and one adult with AMC identified the following important and essential items to be included in the newly developed outcome measure (i.e., picking up an object, eating, reaching mouth, getting out of bed, using spoon/fork, putting on pants, dressing (donning and doffing coat), brushing teeth, and ability to participate in social games with family members. These findings supported the rationale that the outcome measure should include domains of the ICF surrounding Body Functions, Body Structures, and Activities but should also encompass the Participation domain. In the third study, the initial steps required to develop the preliminary version of the SHAPE-UP were carried out. In addition, a three-round survey was disseminated to experts in the field of AMC. This process was iterative in nature as the SHAPE-UP was adapted and modified to reflect the comments and results found throughout the survey process.

In the final study (Chapter 6 – Manuscript 3) the SHAPE-UP was administered to 92 participants across six collaborative sites. Based on the results of the Rasch analysis, the SHAPE-

UP was shortened to a seven-task performance-based outcome measure with the Task Completion scale modified to adopt a 3-point scoring scale instead of a 5-point scoring. Initial psychometric evaluation using Rasch analysis provided evidence of content and construct validity as well as reliability. However, further testing is needed to ensure the unidimensionality of the transformed version of the SHAPE-UP, as well as the collinearity found within items.

By engaging stakeholders and conducting a comprehensive series of studies, the SHAPE-UP was developed and has shown rigorous modern psychometric properties. This measure will not only contribute to improving the quality of care for children and youth with AMC, but also demonstrated the importance of considering participation and personal factors in rehabilitation.

7.2 Stakeholder Engagement

7.2.1 Patient- and family-centered care in health and clinical practice

Family-centered care is broadly defined as "an approach to the planning, delivery, and evaluation of healthcare that is grounded in mutually beneficial partnerships among healthcare providers, patients, and families" [84,85]. The related literature is characterized by terms such as partnership, collaboration, and families as experts to describe the process of care delivery [86]. In pediatrics, the term patient-centered care is often used interchangeably with family-centered care in pediatrics, as the language used to define "patient" refers to "any individual or group with lived experience of a health or health systems issue, including family members, caregivers, and the organizations that are involved with the population of interest" [87–89]. A consensus definition of patient and family-centered care practices and actions has not yet been achieved; however, many organizations have agreed on certain principles that should be included. These are respect and dignity, information sharing, participation, and collaboration [85,86,90,91].

Today, many clinicians working in the pediatric domain organize their clinical practice around the values of patient and family-centered care [91]. Patients and their families are now the focus of the healthcare system as recipients of its services, programs, and delivery approaches. This ensures that services, deliveries, and decisions are made in a collaborative manner to enable best practice and evidence-based interventions to promote health outcomes [92]. Patient and family-centered care is changing the way hospitals provide patient care and recent studies have found significant relationships between patient and family-centered care and

enhanced clinical outcomes, such as patient satisfaction and self-management [93,94]. A systematic review seeking to assess the effects of family-centred models of care for children when compared to standard models of care determined that a patient and family-centered care model had a positive effect on the adequacy of children's care, parental satisfaction, and costs to society [84].

7.2.2 What is patient engagement in health research?

Patient and family-centered care has become the gold standard of healthcare practice in Canada, however, patient engagement is considered a key precursor to quality patient and family-centered care [95]. In the last 10 years, patient engagement in health research has emerged as the next evolution in healthcare delivery. This has allowed for an increased opportunity to involve patients in decision-making related to health research while improving health outcomes [95–97]. Similarly, patient engagement has progressively become accepted and valued among healthcare professionals. Therefore, the number of terms and definitions used to describe similar activities across various institutional settings has also increased exponentially. The terms and definitions have been tailored to different health disciplines, administrations, various countries, and patients themselves [98]. However, Gallivan et al. (2012) reported that demystifying what is meant by patient engagement should be addressed in order to reduce misunderstandings. This can be undertaken by outlining a clear purpose through goals and objectives and determining roles and responsibilities of stakeholders [98]. Additionally, Domecq et al. (2014) conducted a systematic review that determined that an overarching concern of researchers and patients was that patient engagement may become tokenistic resulting in devaluated patients' input [99]. Therefore, it becomes important for all stakeholders involved to have the same expectations in order to avoid potential barriers and achieve meaningful and successful patient engagement [100].

7.2.3 Who are the stakeholders involved in patient engagement in health research?

Based on literature search conducted by Deverka et al. (2012), stakeholders are defined as "individuals, organizations, or communities that have a direct interest in the process and outcomes of a project, research, or policy endeavor" [101]. This broad stakeholder definition was further developed to include stakeholder categories (i.e., patients and consumers, clinicians, healthcare providers, payers and purchasers, policymakers and regulators, life sciences industry,

researchers, and research funders) that should be considered in patient engagement in health research [101]. However, these categories were further adapted to yield the seven main stakeholder categories: patients, healthcare professionals, policymakers, payers, industry, researchers, and research funders [100]. Boudes et al. (2018) created a stakeholder expectation matrix that provides stakeholders' views of relationships, roles, goals, and responsibilities [100]. Therefore, Boudes et al. (2018) determined that patients and their families "promote opportunities for engagement, are informed and actively involved"; healthcare professionals provide "clinical management and patient education"; researchers "understand patient priorities and ensure value of patient input" [100].

7.2.4 Patient engagement in children and youth

Applicability to children and youth is a crucial aspect of patient engagement in general. In 2001, a subcategory of patient engagement known as youth engagement emerged [102]. It was determined that when youth participate in decision-making, they become engaged in the life of their communities [103]. Youth engagement recognizes young people's right to participate in decisions that impact them and acknowledge the great skills and strengths they're capable of contributing [104]. It establishes young people as valued stakeholders into creating effective and inclusive policies, programs, and environments [104]. The WHO (2015) states that adolescents have unique needs that should be treated independently from younger and older patient populations [105]. Through engagement, youth gain a sense of empowerment and form healthy connections to others, which are reflected in the reduction of risky behaviors and increases positive activities [106]. Engaging patients and caregivers in the clinical research process not only provides valuable insight, but may allow researchers to improve their clinical studies through increased enrollment, better participant retention, and ultimately generate more significant outcomes [106]. In addition to the social benefits of these behavioral changes, the community gains through the energy, ideas, and values that youth bring to organizations [103]. Although steps are being made to integrate the patient voice in research, there remains a lack of pediatric specific engagement in clinical research [106,107]. In 2016, the Strategy for Patient-Oriented Research also recognized the importance of pediatric patient-engagement and funded the CHILD-BRIGHT Network, which is a Canadian research network aimed at improving the lives of children with brain-based development disabilities [108,109]. The CHILD-BRIGHT Network uses "family and child-focused approaches to create novel interventions, promote

health outcomes, and deliver responsive and supportive services by meaningfully engaging key stakeholders while paying careful attention to seek the voices of those who may experience greater health care needs due to socio-demographic factors, or those whose voices are insufficiently heard" [108,109]. CHILD-BRIGHT has developed the Public and Patient Engagement Evaluation Tool (PPEET), a standardized survey that evaluates patient engagement processes and impacts based on four sections (i.e., communication, sharing views and perspectives, impacts and influence of engagement initiative, final thoughts/satisfaction). The patient partners' responses on the PPEET ranged between "Agree" to "Strongly Agree" 67-100% for all four sections, with youth having 100% of responses "Agree" or higher for every question on the PPEET [110].

In summary, the importance of engaging youth and caregivers as well as clinician experts in the research on AMC cannot be overstated. Drawing on the literature identifying the various needs of stakeholders in the AMC population, their unique perspectives and lived experiences are invaluable assets in advancing the understanding of this group of rare conditions and improving patient outcomes [68,70]. By actively participating in research initiatives at all levels, stakeholders can help shape the future of healthcare, ensuring that patient and family-centered care remains at the core of everything researchers do.

7.3 Clinical Implications

The innovation in the methodology used to develop the SHAPE-UP is three-fold: engagement of stakeholders (i.e., patients and clinical experts) in the process, tailoring of the choice of items and measurement scale to the needs of the end users (i.e., patients, caregivers, clinicians), and use of sophisticated statistical techniques (i.e., Rasch analysis). The SHAPE-UP provides clinicians with an outcome measure that will impartially characterize and document the functional status of the UE in children and youth with AMC. The development of the measure addresses the uniqueness with which a child with AMC performs a task, as well as their ability to complete tasks of daily living and participation, which was lacking in generic outcome measures. This is a particularly notable contribution given the limited applicability of standardized outcome measures in the context of rare diseases. Considering the heterogeneity of AMC, this novel outcome measure needs to contain a wide spectrum of items that span the complete range of item difficulty, from the easiest task accomplished by the most severely affected individuals to the most

difficult ones completed only by those with mild impairment [77]. The tasks included in the SHAPE-UP elicit both unilateral and bilateral activities as well as account for UE development based on appropriate age groups [77,111]. Additionally, through a multisite collaboration, clinicians from all over North America will be able to use an AMC-specific outcome measure to evaluate children and youth in order to improve treatment selection, functional outcomes and overall quality of life.

7.4 Strengths and Limitations

7.4.1 Strengths

This dissertation encompasses four fundamental strengths that collectively contribute to its significance and impact. Firstly, the development of the SHAPE-UP marks an innovative achievement, representing the first-ever AMC-specific UE outcome measure. This ground-breaking development will elevate the field at both the clinical and research levels. Determining the outcomes of treatment can inform the timing of surgical and rehabilitative interventions on large samples that can be evaluated prospectively, rather than retrospectively as is often the case with rare diseases.

Second, the successful completion of this doctoral thesis underscores the power of collaboration, uniting over 40 research contributors across North America. Our extensive collaborative effort has produced multiple benefits such as fostering strengthened partnerships within the Shriners organization, enriching our understanding of AMC rehabilitation, and fostering the dissemination of knowledge that will in turn translate into improved patient outcomes.

Third, stakeholder engagement was at the heart of the research. Pediatric rare diseases are different from other common chronic health conditions because of the amount of information available. With other chronic pediatric health conditions (e.g., cerebral palsy or spina bifida), the level of public awareness is higher and information is available on rehabilitation and interventions, the prognosis with treatment, and management of the health condition. In rare diseases, the scientific community needs to help patients and families by integrating the firsthand experiences of children and youth, caregivers, and clinicians. This approach aimed to bridge the gap between research insights and practical applications.

Finally, the development of the SHAPE-UP is grounded in modern measurement theory (i.e., Rasch analysis) which is increasingly recognized for its utility as an essential and robust approach to outcome measure development and psychometric validation [112].

In summary, the dissertation's strengths lie in the development of a novel outcome measure, expansive collaborative efforts, stakeholder engagement, and methodological rigor. Collectively, these elements have made a significant and impactful contribution to the field of AMC, clinical practice, and patient outcomes.

7.4.2 Limitations

In addition to the specific limitations described in the individual manuscripts, two main limitations apply to this dissertation on a broader level.

First, due to the heterogeneity and varying clinical presentation of AMC, there is no uniform consensus regarding the classification of AMC and diagnosis of AMC. Consequently, in the development of the SHAPE-UP; it was imperative to develop tasks that were broad enough to accommodate the heterogeneous nature of AMC, while retaining applicability to the majority of children and youth affected. The establishment of a comprehensive classification system holds the potential to facilitate the development of a tailored version of the SHAPE-UP for distinct subgroups within the classification.

Second, in the study of rare diseases, small sample sizes are inevitable. Similarly, to the manuscripts included in this dissertation, in a study with investigating the incidence of thyroid dysfunction among patients with alkaptonuria, a cohort of 125 patients were recruited. While this is by no means a generous sample size, the cohort was the largest of its kind for patients with alkaptonuria [113]. Furthermore, researchers must also be aware of the analytic challenges that arise from studying rare diseases, including the extent to which the available data can be viewed as representative of the entire population of patients with the condition and whether there is sufficient statistical power to draw definitive conclusions [114]. However, one of the promising advantages to using Rasch analysis, is the generalizability across samples and items, missing data, and identification of poorly functioning items. This was indeed confirmed by Waterbury (2019), who stated "the Rasch model can handle varying amounts of missing data" [115]. Additionally, in a study evaluating the Rasch analysis results using small sample sizes, the results suggest that Rasch modelling on small sample sizes ($n<250$) should be used for exploratory purposes where researchers not make any definitive decisions about deleting items or collapsing response

categories [116]. Therefore, increasing the dissemination of information on rare diseases research could enhance awareness and potentially facilitate new partnerships among different institutions, resulting in small data becoming bigger.

7.5 Directions for Future Work

7.5.1 Psychometrics properties

Following the development of the SHAPE-UP, there remains a critical need for rigorous validation. Considering that evidence on the effectiveness of interventions in AMC is lacking, thorough measurement research is required to advance the quality care provided to children with AMC and their families. Evaluating the outcomes of treatment can inform the timing of specific interventions, which then facilitates the evaluation of treatment effectiveness. Specifically, outcomes of surgical and clinical (e.g., bracing, casting) interventions on large samples can be evaluated prospectively, rather than retrospectively. This data can inform clinicians and researchers as to the suitable timing of specific interventions to improve functional outcomes, minimize morbidity, and promote quality of life, thus promoting treatment advances in the field.

A study in 2015 has shown that one way to facilitate the uptake of newly developed outcome measures is to demonstrate their utility to stakeholders [117]. Establishing the psychometric properties of a newly established measure is essential to demonstrate that the construct is stable over time for the same examiner and among different examiners.

A multisite 3-year clinical grant funded by Shriners' Children (2024-2026; PI Dahan-Oliel) will evaluate inter, intra and test retest reliability, validity and responsiveness among children with AMC across the same six hospital sites as Study 4 (Manuscript 3). The classical reliability and validity of the SHAPE-UP will be established by determining the inter-rater and intra-rater reliability, test-retest while the convergent and discriminant validity will be evaluated across the six participating sites to ensure the measure is stable over time, among raters, and measures what it is intended do. Furthermore, determining the ability of the measure to capture meaningful clinical change (i.e., responsiveness) will enable clinicians evaluate the effectiveness of treatment. In addition to the quantitative psychometrics evaluations, a qualitative descriptive study design comprised of individual interviews will be employed to describe the perspectives of stakeholders regarding the utility of the SHAPE-UP in guiding clinical decision-making in this population.

The psychometric analysis of a reliable, valid, responsive and useful outcome measure to evaluate the UE in children with AMC will allow tailored assessments in AMC leading to evidence-based interventions.

7.5.2 Knowledge Translation

Knowledge translation is defined by CIHR as "A dynamic and iterative process that includes the synthesis, dissemination, exchange and ethically sound application of knowledge to improve health, provide more effective health services and products, and strengthen the health care system" [118,119]. As reported earlier, there has been a growing recognition with regards to the need for including patients and members of the public within research processes [120]. The most utilized framework to promote the application of research and implementation is the Knowledge-to-Action (KTA) Framework (Figure 7-1) developed by Graham et al. (2006) [118,120–124].

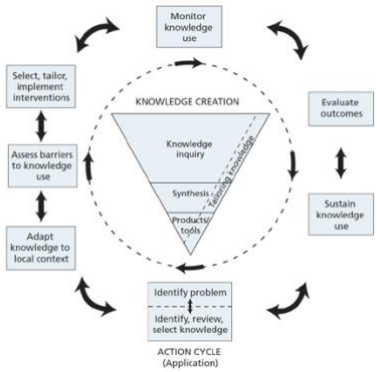

Figure 7-1 Knowledge-to-Action Framework

Using the KTA Framework assures conceptual clarity, objective definitions of ambiguous terminology, and ultimately provides an exchange of knowledge between relevant stakeholders that results in action. For example, during the planned 4th International Symposium for Arthrogryposis (Shriners Hospital for Children – Canada, Montreal, in September 2024), the development team of the SHAPE-UP will prepare a training session on how to 1) properly set up the video camera to ensure all tasks and subtasks are visible for scoring, 2) administer the measure using the toolkit provided, 3) appropriately score the child with AMC performing the task using the Task Completion scale and the Analysis of Joint Motion and Position. There will also be mock scoring scenarios presented to the audience to ensure all participants have a correct understanding and application of the scoring tables.

In addition to the training session, more traditional methods of sharing the SHAPE-UP will be disseminated. The various studies comprising the development of the SHAPE-UP have been submitted/published in peer-reviewed journals. Abstracts and posters have been submitted and presented at local, national, and international conferences. The SHAPE-UP will also be made available to share with rehabilitation and hospital settings worldwide through an international consortium for AMC that my supervisor has developed in 2021. Flyers and information shared via social media platforms and through the AMC support groups online (i.e., Arthrogryposis Multiplex Congenita Support Inc (AMCSI) website). The dissemination of the SHAPE-UP will be a multifaceted approach to ensure that the outcome measure and knowledge established by the SHAPE-UP development team will reach and benefit AMC communities worldwide.

7.6 Concluding Statement

In conclusion, this thesis dissertation makes an original and valuable contribution to the field of measurement development, pediatric rehabilitation and rare diseases. The three manuscripts presented in this thesis 1) identify the constraints of the ICF Framework and underscore the significance of the Participation domain, 2) describe the development of the preliminary version the UE AMC-specific pediatric outcome measure using stakeholder engagement, and 3) demonstrate the fit to the Rasch model of the SHAPE-UP. It is clear that robust and rigorous PBOM have the potential to transform clinical practice by 1) empowering clinicians in their mission to provide the best possible care and improve patient outcomes, and 2) inform researchers and decision-makers on the benefits of stakeholder engagement at all levels of medical care.

In the course of my doctoral training and thesis research, I have established a commitment to methodological rigour and originality, developed a novel outcome measure in the field of AMC, and revealed the benefits of stakeholder engagement in childhood disability research.

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Appendices

Appendix 1. Manuscript 1 - Table 4. Linking rules data extraction table for 32 outcome measures.

1. Name of instrument	2. Item aka Verbatim health information	3. Perspective	4. Response options	5. Classification of response	6. Main meaningful concept	7. Additional meaningful concept	8. ICF Category: Main meaningful concept	9. ICF category: Other meaningful concept	10. Annotation
Assisting Hand Assessment (AHA)	*AH Initiation/usage*								
	Touches	Capacity	N/A	N/A	touch		d1201 - Touching		
	Holds	Capacity	N/A	N/A	hold		d4401 - Grasping		
	Initiates use	Capacity	N/A	N/A	initiating		d2100 - Undertaking a simple task		
	Attends assisting hand	Capacity	N/A	N/A	attends assisting hand		d4458 - Hand and arm use, other specified		
	Stabilizes by weight	Capacity	N/A	N/A	stabilizes		b7158 - Stability of joint function, other specified		general stability
	Arm use								
	Proceeds	Capacity	N/A	N/A	proceeds		d2100 - Undertaking a simple task		
	Uses assisting hand	Capacity	N/A	N/A	uses assisting hand		d4458 - Hand and arm use, other specified		
	Strategies	Capacity	N/A	N/A	strategies		d1759 - Solving problems, unspecified		general problem solving
	Grip-release functions								
	Stabilizes by grip	Capacity	N/A	N/A	grip	stability	d4401 - Grasping	b7158 - Stability of	general stability

								joint function, other specified	
	Hand choice	Capacity	N/A	N/A	hand choice		b7602 - Coordination of voluntary movements		
	Coordinates	Capacity	N/A	N/A	coordination		b7602 - Coordination of voluntary movements		
	Reaches	Capacity	N/A	N/A	reach		d4452 - Reaching		
	Varies fingers	Capacity	N/A	N/A	finger movement		b7600 - Control of simple voluntary movements		
	Fine motor adjustments								
	Grip types	Capacity	N/A	N/A	grip		d4401 - Grasping		
	Releases	Capacity	N/A	N/A	release		d4403 - Releasing		
	Grasp hold of	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Flows	Capacity	N/A	N/A	flow		d4409 - Fine hand use, unspecified		flowing
	Coordination ability								
	Calibrates	Capacity	N/A	N/A	calibration		d4458 - Hand and arm use, other specified		
	Varies arm position	Capacity	N/A	N/A	arm coordination		b7602 - Coordination of voluntary movements		
	Performance pace								

	Varies grips	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Adjusts object	Capacity	N/A	N/A	adjusts object		d4402 - Manipulating		
	Manipulates	Capacity	N/A	N/A	manipulates		d4402 - Manipulating		
Melbourne assessment of unilateral upper limb function-2(MAUUL-2)	Reach forward	Capacity	N/A	N/A	reach		d4452 - Reaching		
	Reach sideways to elevated position	Capacity	N/A	N/A	reach		d4452 - Reaching		
	Grasp of crayon	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Drawing grasp	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Release of crayon	Capacity	N/A	N/A	releasing		d4403 - Releasing		
	Grasp of pellet	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Release of pellet	Capacity	N/A	N/A	releasing		d4403 - Releasing		
	Manipulation	Capacity	N/A	N/A	manipulation		d4402 - Manipulating		
	Pointing	Capacity	N/A	N/A	pointing		b7600 - Control of simple voluntary movements		
	Reach to brush from forehead to back of neck	Capacity	N/A	N/A	grooming		d520 - Caring for body parts		
	Palm to bottom -> simulating wiping	Capacity	N/A	N/A	wiping		d530 - Toileting		
	Pronation/supination	Capacity	N/A	N/A	pronation/supination		b7100 - Mobility of a single joint		
	Reach to opposite shoulder	Capacity	N/A	N/A	reach to opposite shoulder		b7602 - Coordination of voluntary movements		
	Hand to mouth and down	Capacity	N/A	N/A	hand to mouth and down		b7602 - Coordination of		

							voluntary movements		
Jebsen-Taylor Hand Function (JTHF)	Writing a short sentence (24 letters, 3rd grade reading difficulty)	Capacity	N/A	N/A	writing		d1700 - Using general skills and strategies of the writing process		
	Turning over a 3x5 inch card	Capacity	N/A	N/A	turning card		d4402 - Manipulating		
	Picking up small common objects	Capacity	N/A	N/A	picking up small object		d4400 - Picking up		
	Simulated feeding	Capacity	N/A	N/A	feeding		d550 - Eating		
	Stacking checkers	Capacity	N/A	N/A	stacking		d4402 - Manipulating		
	Picking up large light cans	Capacity	N/A	N/A	picking up light object		d4408 - Fine hand use, other specified		large object
	Picking up large heavy cans	Capacity	N/A	N/A	picking up heavy object		d4408 - Fine hand use, other specified		large object
Quality of Upper Extremity Skills Test (QUEST)	*Dissociated Movements*								
	Shoulder								
	Flexion	Capacity	N/A	N/A	flexion		b7600 - Control of simple voluntary movements		
	Flexion with fingers extended	Capacity	N/A	N/A	flexion with fingers extended		b7601 - Control of complex voluntary movements		
	Abduction	Capacity	N/A	N/A	abduction		b7600 - Control of simple voluntary movements		
	Abduction with fingers extended	Capacity	N/A	N/A	abduction with fingers extended		b7601 - Control of complex voluntary movements		
	Elbow								

	Flexion with supination	Capacity	N/A	N/A	flexion with supination		b7601 - Control of complex voluntary movements		
	Extension with supination	Capacity	N/A	N/A	extension with supination		b7601 - Control of complex voluntary movements		
	Flexion with pronation	Capacity	N/A	N/A	flexion with pronation		b7601 - Control of complex voluntary movements		
	Extension with pronation	Capacity	N/A	N/A	extension with pronation		b7601 - Control of complex voluntary movements		
	Wrist								
	Extension with elbow extension	Capacity	N/A	N/A	extension with elbow extension		b7601 - Control of complex voluntary movements		
	Extension with elbow flexion	Capacity	N/A	N/A	extension with elbow flexion		b7601 - Control of complex voluntary movements		
	Extension with pronation	Capacity	N/A	N/A	extension with pronation		b7601 - Control of complex voluntary movements		
	Extension with supination	Capacity	N/A	N/A	extension with supination		b7601 - Control of complex voluntary movements		
	Flexion with supination	Capacity	N/A	N/A	flexion with supination		b7601 - Control of complex voluntary movements		
	Independent movement								
	Fingers	Capacity	N/A	N/A	fingers		b7600 - Control of simple		

							voluntary movements		
	Thumb	Capacity	N/A	N/A	thumb		b7600 - Control of simple voluntary movements		
	Arm position during grasp/release								
	Grasp using thumb	Capacity	N/A	N/A	grasp using thumb		d4401 - Grasping		
	Grasp using palm	Capacity	N/A	N/A	grasp using palm		d4401 - Grasping		
	Release from thumb and fingers	Capacity	N/A	N/A	releasing thumb and fingers		d4403 - Releasing		
	Release from palm	Capacity	N/A	N/A	release palm		d4403 - Releasing		
	Sitting Posture during Grasp								
	Head	Capacity	N/A	N/A	posture		b7601 - Control of complex voluntary movements		
	Trunk	Capacity	N/A	N/A	body posture		b7601 - Control of complex voluntary movements		
	Shoulders	Capacity	N/A	N/A	posture		b7601 - Control of complex voluntary movements		
	Grasp								
	Grasp of 1" cube	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Grasp of cereal	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Grasp of pencil/crayon	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Weight Bearing								

	Weight bearing in prone	Capacity	N/A	N/A	weight bearing		b7603 - Supportive functions of arm or leg		
	Weight bearing in prone with reach	Capacity	N/A	N/A	weight bearing		b7603 - Supportive functions of arm or leg		
	Weight bearing in sitting with hands forward	Capacity	N/A	N/A	weight bearing		b7603 - Supportive functions of arm or leg		
	Weight bearing in sitting with hands by side	Capacity	N/A	N/A	weight bearing		b7603 - Supportive functions of arm or leg		
	Weight bearing in sitting with hands behind	Capacity	N/A	N/A	weight bearing		b7603 - Supportive functions of arm or leg		
	Protective Extension								
	Protective extension - forward	Capacity	N/A	N/A	protective extension		b755 - Involuntary movement reaction functions		
	Protective extension - side	Capacity	N/A	N/A	protective extension		b755 - Involuntary movement reaction functions		
	Protective extension - backward	Capacity	N/A	N/A	protective extension		b755 - Involuntary movement reaction functions		
Box and Blocks	*Dominant Hand*								
	I want to see how quickly you can pick up one block	Capacity	N/A	N/A	pick up block, carry block, drop block, fingertips		d4400 - Picking up; d4301 - Carrying in the		

	at a time with your right (or left) hand [examiners points to the hand]. Carry it to the other side of the box and drop it. Make sure your fingertips cross the partition. Watch me while I show you. If you pick up two blocks at a time, they will count as one. If you drop one on the floor or table after you have carried it across, it will still be counted, so do not waste time picking it up. If you toss the blocks without your fingertips crossing the partition, they will not be counted. Before you start, you will have a chance to practice for 15 seconds. Do you have any questions? Place your hands on the sides of the box. When it is time to start, I will "ready" and then "go".				across partition, toss blocks, place hand on side of box		hands; d4403 - Releasing ; b7600 - Control of simple voluntary movements ; d4454 - Throwing; b7600 - Control of simple voluntary movements		
	Non-dominant hand								

	Now you are to do the same thing with your left (or right) hand. First you can practice. Put your hands on the sides of the box as before. Pick up one block at a time with your hand, and drop it on the other side of the box. Ready. Go.	Capacity	N/A	N/A	put hands on sides of box; pick up one block; drop block on other side of box		b7600 - Control of simple voluntary movements; d4400 - Picking up; d4403 - Releasing		
Bruininks-Oseretsky Test of Motor Proficiency (BOT-2)	*Fine motor Precision Domain*								
	Filling in shapes - circle	Capacity	N/A	N/A	drawing or colouring		d4408 - Fine hand use, other specified		colouring
	Filling in shapes - star	Capacity	N/A	N/A	filling in shape		d4408 - Fine hand use, other specified		colouring in
	Drawing lines through paths - crooked	Capacity	N/A	N/A	drawing crooked lines		d4408 - Fine hand use, other specified		drawing
	Drawing lines through paths - curved	Capacity	N/A	N/A	drawing curved lines		d4408 - Fine hand use, other specified		drawing
	Connecting dots	Capacity	N/A	N/A	connecting dots		d4408 - Fine hand use, other specified		connecting dots
	Folding paper	Capacity	N/A	N/A	folding paper		d4402 - Manipulating		
	Cutting out a circle	Capacity	N/A	N/A	cutting a circle		d4402 - Manipulating		
	Fine motor Integration Domain								
	Copying a circle	Capacity	N/A	N/A	copying a circle		d4408 - Fine hand use, other specified		drawing

	Copying a square	Capacity	N/A	N/A	coping a square		d4408 - Fine hand use, other specified		drawing
	Copying overlapping circles	Capacity	N/A	N/A	copying overlapping circles		d4408 - Fine hand use, other specified		drawing
	Copying a wavy line	Capacity	N/A	N/A	copying a wavy line		d4408 - Fine hand use, other specified		drawing
	Copying a triangle	Capacity	N/A	N/A	copying a triangle		d4408 - Fine hand use, other specified		drawing
	Copying a diamond	Capacity	N/A	N/A	copying a diamond		d4408 - Fine hand use, other specified		drawing
	Copying a star	Capacity	N/A	N/A	copying a star		d4408 - Fine hand use, other specified		drawing
	Copying overlapping pencils	Capacity	N/A	N/A	copying overlapping pencils		d4408 - Fine hand use, other specified		drawing
	Manual Dexterity Domain								
	Making dots in circles	Capacity	N/A	N/A	making dots in circles		d4408 - Fine hand use, other specified		making dots
	Transferring pennies	Capacity	N/A	N/A	transferring pennies		d4402 - Manipulating		
	Placing pegs into a pegboard	Capacity	N/A	N/A	placing pegs into a peg board		d4402 - Manipulating		
	Sorting cards	Capacity	N/A	N/A	sorting cards		d4402 - Manipulating		
	Stringing blocks	Capacity	N/A	N/A	stringing blocks		d4402 - Manipulating		
	Upper limb coordination Domain								
	Dropping and catching a ball - both hands	Capacity	N/A	N/A	dropping ball	catching ball	d4403 - Releasing	d4455 - Catching	

	catching a tossed ball - both hands	Capacity	N/A	N/A	catching a ball with both hands		d4455 - Catching		
	dropping and catching a ball - one hand	Capacity	N/A	N/A	dropping ball with one hand	catching	d4403 - Releasing	d4455 - Catching	
	catching a tossed ball - one hand	Capacity	N/A	N/A	catching a ball with one hand		d4455 - Catching		dribbling
	Dribbling a ball - one hand	Capacity	N/A	N/A	dribbling a ball with one hand		d4458 - Hand and arm use, other specified		dribbling
	Dribbling a ball - alternating hands	Capacity	N/A	N/A	dribbling a ball with alternating hands		d4458 - Hand and arm use, other specified		
	throwing a ball at a target	Capacity	N/A	N/A	throwing a ball at a target		d4454 - Throwing		
Movement Assessment Battery for Children - 2	*Age band 1*								
	Manual Dexterity								
	Posting coins preferred hand	Capacity	N/A	N/A	posting coins		d4402 - Manipulating		
	posting coins non-preferred hand	Capacity	N/A	N/A	posting coins		d4402 - Manipulating		
	threading beads	Capacity	N/A	N/A	threading beads		d4402 - Manipulating		
	drawing trail 1	Capacity	N/A	N/A	drawing trail		d4408 - Fine hand use, other specified		drawing trail
	Aiming and catching								
	catching beanbag	Capacity	N/A	N/A	catching		d4455 - Catching		
	throwing beanbag onto mat	Capacity	N/A	N/A	throwing		d4454 - Throwing		
	Age band 2								
	Manual Dexterity								
	placing pegs preferred hand	Capacity	N/A	N/A	placing pegs		d4402 - Manipulating		

	placing pegs non-preferred hand	Capacity	N/A	N/A	placing pegs		d4402 - Manipulating		
	threading lace	Capacity	N/A	N/A	threading lace		d4402 - Manipulating		
	drawing trail 2	Capacity	N/A	N/A	drawing trail		d4408 - Fine hand use, other specified		drawing trail
	Aiming and catching								
	catching with two hands	Capacity	N/A	N/A	catching		d4455 - Catching		
	throwing beanbag onto mat	Capacity	N/A	N/A	throwing		d4454 - Throwing		
	Age band 3								
	Manual Dexterity								
	Turning pegs preferred hand	Capacity	N/A	N/A	turning pegs		d4453 - Turning or twisting the hands or arms		
	Turning pegs non-preferred hand	Capacity	N/A	N/A	turning pegs		d4453 - Turning or twisting the hands or arms		
	triangle with nuts and bolts	Capacity	N/A	N/A	triangle nuts and bolts		d4402 - Manipulating		
	drawing trail 3	Capacity	N/A	N/A	drawing trail		d4408 - Fine hand use, other specified		drawing trail
	Aiming and catching								
	Catching with 1 hand best hand	Capacity	N/A	N/A	catching		d4455 - Catching		
	catching with 1 hand other hand	Capacity	N/A	N/A	catching		d4455 - Catching		
	throwing at wall target	Capacity	N/A	N/A	throwing		d4454 - Throwing		
Functional Independence Measure	Eating	Performance	N/A	N/A	eating		d550 - Eating		
	Grooming	Performance	N/A	N/A	grooming		d520 - Caring for body parts		

for Children (WeeFIM)	Bathing	Performance	N/A	N/A	bathing		d510 - Washing oneself		
	Dressing - upper	Performance	N/A	N/A	dressing upper body		d540 - Dressing		
	Dressing - lower	Performance	N/A	N/A	dressing lower body		d540 - Dressing		
	Toileting	Performance	N/A	N/A	toileting		d530 - Toileting		
Peabody Developmental Motor Scales (PDMS) -2	*Object Manipulation*								
	Catching ball [Sitting legs spread apart facing you, you and child sitting 3 ft apart] (Roll ball from between your legs to child "Catch the ball")	Capacity	N/A	N/A	catching		d4455 - Catching		
	Rolling ball [sitting legs spread apart facing you, you and child sitting 3 ft apart] (Roll ball from between your legs to child. Place ball on floor between child's knees "Roll the ball to me")	Capacity	N/A	N/A	rolling		d4458 - Hand and arm use, other specified		rolling
	Flinging ball [standing in an open area] (give tennis ball to child and stand 5 ft away. Extend your hands to child "Throw the ball to me"	Capacity	N/A	N/A	flinging		d4454 - Throwing		
	Throwing ball [standing in an	Capacity	N/A	N/A	throwing		d4454 - Throwing		

	open area] (give tennis ball to child and stand 5 ft away "throw the ball to me"								
	Throwing ball overhand [standing in an open area] (demonstrate throwing tennis ball overhand at least 3 ft forward. Give the ball to child "throw the ball as far as you can"	Capacity	N/A	N/A	throwing	as far as you can	d4454 - Throwing	b7301 - Power of muscles of one limb	
	Throwing ball underhand [standing in an open area] (demonstrate throwing a tennis ball underhand at least 5 ft. give ball to child "throw the ball as far as you can"	Capacity	N/A	N/A	throwing	as far as you can	d4454 - Throwing	b7301 - Power of muscles of one limb	
	Catching ball [standing in an open area] stand 5 ft. in front of child "Catch the ball" Toss ball so that it arrives at chest height, contacting child's outstretched arms	Capacity	N/A	N/A	catching		d4455 - Catching		
	Throwing ball overhand [standing in an open area] demonstrate	Capacity	N/A	N/A	throwing		d4454 - Throwing		

	throwing tennis ball overhand at least 7 ft. give ball to child stand 8 ft away "throw me the ball"								
	Throwing ball underhand [standing in an open area] Demonstrates throwing the tennis ball underhand at least 7 ft. forward. Give the ball to child stand 8 ft. away "throw me the ball"	Capacity	N/A	N/A	throwing		d4454 - Throwing		
	Catching ball [standing in an open area] Stand 5 ft. in front of child "Catch the ball" Toss ball so that it arrives at chest height, contacting child's outstretched arms	Capacity	N/A	N/A	catching		d4455 - Catching		
	Throwing ball - overhand [standing in an open area] Demonstrate throwing tennis ball overhand at least 10 ft. give ball to child. Stand 11 ft. away "Throw the ball as far as you can"	Capacity	N/A	N/A	throwing	as far as you can	d4454 - Throwing	b7301 - Power of muscles of one limb	
	Hitting target - underhand	Capacity	N/A	N/A	throwing	hitting target	d4454 - Throwing	b7602 - Coordinatio	

	[standing 5ft. From wall] From 5 ft. away toss tennis ball underhand to 2 ft. target taped on wall (2ft. Above floor) "throw the ball and hit the target like I did"							n of voluntary movements	
	Catching ball [standing in an open area] Stand 5ft. In front of child "Catch the ball" Toss ball so that it arrives at chest height	Capacity	N/A	N/A	catching		d4455 - Catching		
	Hitting target - overhand [standing 5 ft. from wall] From 5 ft. away toss tennis ball twice overhand to 2 ft. target taped on wall (2 ft. above floor) "throw the ball and hit the target like I did"	Capacity	N/A	N/A	throwing	hitting target	d4454 - Throwing	b7602 - Coordination of voluntary movements	
	Throwing ball - underhand [standing in an open area] Demonstrate throwing tennis ball underhand at least 10ft. Give ball to child. Stand about 12 ft. away "Throw the ball as far as you can"	Capacity	N/A	N/A	throwing	as far as you can	d4454 - Throwing	b7301 - Power of muscles of one limb	

	Hitting target - overhand [standing 12 ft. from wall] From 12 ft. away toss tennis ball overhand to 2 ft. target taped on wall (2 ft. above floor) "throw the ball and hit the target like I did"	Capacity	N/A	N/A	throwing	hitting target	d4454 - Throwing	b7602 - Coordination of voluntary movements	
	Bouncing ball [standing 5 ft. from wall] using 1 hand bounce tennis ball so it bounces once and then hits wall. Give ball to child "bounce the ball like I did"	Capacity	N/A	N/A	bouncing		d4458 - Hand and arm use, other specified		bouncing
	Catching ball [standing in an open area] stand 5 ft. in front of child "Catch the ball" toss tennis ball in a 45-degree arc so it arrives at child's hands	Capacity	N/A	N/A	catching		d4455 - Catching		
	Catching bounced ball Bounce tennis ball on floor once and catch it with 1 hand "Bounce and catch the ball like I did"	Capacity	N/A	N/A	catching		d4455 - Catching		
	Grasping								
	Grasping reflex [lying on back]	Capacity	N/A	N/A	grasping reflex		b7502 - Reflexes		

	Stimulate child's palm by inserting your index finger into thumb side of palm						generated by other exteroceptive stimuli		
	Grasping cloth [lying on back] spread washcloth over your forearm place child's hand on top of washcloth	Capacity	N/A	N/A	grasping		d4401 - Grasping		
	Releasing rattle - disappearing reflex [lying on back] place rattle in child's hand. After child holds rattle for 5 seconds observe amount of time before release	Capacity	N/A	N/A	releasing		d4403 - Releasing		
	Grasping rattle [lying on back] Lightly touch child's palm with rattle "Get your rattle"	Capacity	N/A	N/A	grasping		d4401 - Grasping		
	Holding rattle [lying on back] place rattle in child's hand	Capacity	N/A	N/A	holding		d4401 - Grasping		
	Manipulating rattle [lying on back] shake rattle and place it in child's hand "Shake your rattle"	Capacity	N/A	N/A	manipulating		d4402 - Manipulating		
	Grasping rattle [sitting on lap facing table] Place rattle on	Capacity	N/A	N/A	grasping		d4401 - Grasping		

	table within 3 in. of child's hand. "Get your rattle"								
	Pulling string [lying on stomach] place toy on a string so string is at midline between child's hands "Get the toy"	Capacity	N/A	N/A	pulling string		d4450 - Pulling		
	Securing paper [sitting on lap, facing table] place 8.5x11 in. paper within 3 in. of child's hand "Get the paper"	Capacity	N/A	N/A	securing paper		d4402 - Manipulating		
	Grasping cube [sitting on lap facing table] place cube on table within 3in. Of child's hand "Get the block"	Capacity	N/A	N/A	grasping		d4401 - Grasping		
	Grasping cube [sitting on lap facing table] place cube on table within 3 in. of child's hand "Get the block" observe how the child picks up cube	Capacity	N/A	N/A	grasping		d4401 - Grasping		
	Shaking rattle [sitting on lap facing table] Place rattle in child's hand "Shake your rattle"	Capacity	N/A	N/A	shaking		d4402 - Manipulating		

	Shaking rattle [sitting on lap facing table] Shake rattle back and forth through a 90-degree arc 3 times. Place it on table in front of child "Shake the rattle"	Capacity	N/A	N/A	shaking		d4402 - Manipulating		
	Grasping cube [sitting on lap facing table] Place cube on table within 3 in. of child's hand. "Get the block" Observe how child picks up cube	Capacity	N/A	N/A	grasping		d4401 - Grasping		
	Grasping pellets [sitting on lap facing table] Place 2 food pellets on table within child's reach "Get all the food"	Capacity	N/A	N/A	grasping		d4401 - Grasping		
	Manipulating paper [sitting on lap facing table] cut 8.5x11 in. sheet of paper in half. Place half on table "Watch me crumple the paper" Crumple paper in 1 hand. Place other half of paper within 3 in. of child's hand "Crumple paper like I did"	Capacity	N/A	N/A	manipulating		d4402 - Manipulating		

	Grasping pellets [sitting on lap facing table] Place 2 food pellets on table within child's reach "Get all the food"	Capacity	N/A	N/A	grasping		d4401 - Grasping		
	Grasping pellets [sitting on lap facing table] place 2 food pellets on table within child's reach "Get all the food"	Capacity	N/A	N/A	grasping		d4401 - Grasping		
	Grasping cube [sitting on lap facing table] place cube on table within 3 in. of child's hand. "Get the block" Observe how child picks up cube	Capacity	N/A	N/A	grasping		d4401 - Grasping		
	Grasping cube [sitting on lap facing table] place 2 cubes side by side. Pick up both cubes with 1 hand. Place cubes on table "Pick up both blocks with 1 hand like I did"	Capacity	N/A	N/A	grasping		d4401 - Grasping		
	Grasping marker [sitting at table] Place paper and marker by child's hand on table "make a mark"	Capacity	N/A	N/A	grasping		d4401 - Grasping		

	Observe how child holds marker								
	Grasping marker [sitting at table] place paper and marker by child's hand on table "Make a mark" Observe how child holds marker	Capacity	N/A	N/A	grasping		d4401 - Grasping		
	Unbuttoning buttons [sitting at table] Place button strip on table "unbutton these as fast as you can"	Capacity	N/A	N/A	unbuttoning	as fast as you can	d4408 - Fine hand use, other specified	b7602 - Coordination of voluntary movements	unbuttoning
	Buttoning button [sitting at table] Place button strip on table. Unbutton the buttons. Point to an end button "Button and unbutton this one as fast as you can"	Capacity	N/A	N/A	buttoning	as fast as you can	d4408 - Fine hand use, other specified	b7602 - Coordination of voluntary movements	buttoning
	Grasping marker [sitting on table] Place paper and marker by child's hand on table "Make a mark" Observe how child holds marker	Capacity	N/A	N/A	grasping		d4401 - Grasping		
	Touching fingers. At the rate of 1 touch per second, beginning with	Capacity	N/A	N/A	touching fingers	as fast as you can	b7602 - Coordination of voluntary movements	b7602 - Coordination of	

	index finger, touch each finger in succession to thumb "touch like I did as fast as you can"							voluntary movements	
Performance of Upper Limb (PUL)	Thumb key grip	Capacity	N/A	N/A	grip		d4401 - Grasping		
	3-point grip	Capacity	N/A	N/A	grip		d4401 - Grasping		
	finger pinch	Capacity	N/A	N/A	finger pinch		d4401 - Grasping		
	place fingers on diagram	Capacity	N/A	N/A	place finger on diagram		b7602 - Coordination of voluntary movements		
	pick up coins	Capacity	N/A	N/A	pick up		d4400 - Picking up		
	supination	Capacity	N/A	N/A	supination		b7100 - Mobility of a single joint		
	push on a light	Capacity	N/A	N/A	push on a light		d4451 - Pushing		
	tracing a path	Capacity	N/A	N/A	tracing a path		d4408 - Fine hand use, other specified		tracing
	tearing paper	Capacity	N/A	N/A	tearing paper		d4402 - Manipulating		
	remove lid from container	Capacity	N/A	N/A	removing lid		d4453 - Turning or twisting the hands or arms		
	Stack light and heavy cans	Capacity	N/A	N/A	stacking cans	weight component - cans	d4402 - Manipulating	b7301 - Power of muscles of one limb	
	Lift light and heavy cans	Capacity	N/A	N/A	lifting cans	weight component - cans	d4300 - Lifting	b7301 - Power of muscles of one limb	
	moving weights on table (100g,	Capacity	N/A	N/A	moving weights	weight component - cans	d4458 - Hand and arm use, other specified	b7301 - Power of	moving

	200g, 500g, 1000g)							muscles of one limb	
	hand to table	Capacity	N/A	N/A	hand to table		b7602 - Coordination of voluntary movements		
	hand to mouth with and without weights (50g, 200g)	Capacity	N/A	N/A	hand to mouth	weight component	b7602 - Coordination of voluntary movements	b7301 - Power of muscles of one limb	
	lifting upper extremity weights (50g, 200g, 500g, 1000g) at shoulder height	Capacity	N/A	N/A	lifting shoulder height	weight component	d4300 - Lifting	b7301 - Power of muscles of one limb	
	lifting upper extremity weights (50g, 200g, 500g, 1000g) above shoulder height	Capacity	N/A	N/A	lifting above shoulder	weight component	d4300 - Lifting	b7301 - Power of muscles of one limb	
Shriners Hospital for Children Upper Extremity Evaluation (SHUEE)	*Spontaneous functional analysis/dynamic positional analysis*								
	Take the paper money out of the wallet like you normally would	Capacity	N/A	N/A	take the money		d4400 - Picking up		
	Now fold this paper in 1/2 in any fashion	Capacity	N/A	N/A	fold the paper		d4402 - Manipulating		
	I'm going to start tearing this paper and I want you to finish it	Capacity	N/A	N/A	tearing paper		d4402 - Manipulating		
	Now I want you to string these beads	Capacity	N/A	N/A	stringing beads		d4402 - Manipulating		

	Please take the top off of this bottle	Capacity	N/A	N/A	remove bottle top		d4453 - Turning or twisting the hands or arms		
	Now tear this Play-Doh into three pieces	Capacity	N/A	N/A	tearing		d4402 - Manipulating		
	Pretend this is something good to eat and show me how you would cut it	Capacity	N/A	N/A	cutting food		d550 - Eating		
	Now let's stand up. I want you to stand over here. I want you to toss this ball to me and I'll toss it back	Capacity	N/A	N/A	standing; throwing; catching		d4104 - Standing; d4454 - Throwing; d4455 - Catching		
	Let me give you these coins	Capacity	N/A	N/A	handling coins		d4402 - Manipulating		
	Let me give you 5	Capacity	N/A	N/A	high five		b7602 - Coordination of voluntary movements		
	Now I want you to show me how you would eat this. You don't have to eat it if you don't want to. Just take it into your mouth	Capacity	N/A	N/A	Eating		d550 - Eating		
	Can you touch this palm to this ear?	Capacity	N/A	N/A	coordinated movement		b7602 - Coordination of voluntary movements		
	Put this sticker on this ball	Capacity	N/A	N/A	coordinated movement		b7602 - Coordination of voluntary movements		
	Go ahead and put your sock back on for me	Capacity	N/A	N/A	dressing (sock)		d5402 - Putting on footwear		

	Now tie your shoe	Capacity	N/A	N/A	Tying		d5408 - Dressing, other specified		Tying
	I know this is silly but I want you to get down and crawl towards the camera	Capacity	N/A	N/A	Crawl	Get down	d4550 - Crawling	d4108 - Changing basic body position, other specified	Get down
	Grasp								
	Can you take this bead from me?	Capacity	N/A	N/A	pick up		d4400 - Picking up		
	Now can you straighten your wrist and still pick up the bead?	Capacity	N/A	N/A	pick up	straighten wrist	d4400 - Picking up	b7100 - Mobility of a single joint	
	Now can you bring your wrist all the way up and pick up the bead?	Capacity	N/A	N/A	pick up	straighten all the way	d4400 - Picking up	b7100 - Mobility of a single joint	
PURDUE pegboard	*Right hand*								
	"Pick up one pin at a time with your right hand from the right-handed cup. Starting with the top hole, place each pin in the right-handed row. (Leave the pin used for demonstration in the hole.) Now you may insert a few pins for practice. If during the testing time you drop a pin, do not stop to pick it up. Simply	Capacity	N/A	N/A	pick up; place each pin; insert a few pins; pick up another pin; remove practice pins		d4400 - Picking up; d4402 - Manipulating; d4402 - Manipulating; d4400 - Picking up; d4403 - Releasing;		

	continue by picking another pin out of the cup." "Stop. Now take out the practice pins and put them back into the right-handed cup." "When I say 'Begin,' place as many pins as possible in the right-handed row, starting with the top hole. Work as rapidly as you can until I say 'Stop.' Are you ready? Begin." "Stop"								
	Left hand								
	"Pick up one pin at a time with your left hand from the left-handed cup. Place each pin in the left-handed row, starting with the top hole. You may insert a few pins for practice." "Stop. Now take out the practice pins, and put them back into the left-handed cup." "When I say 'Begin,' place as many pins as possible in the left-handed row,	Capacity	N/A	N/A	pick up; place each pin; insert a few pins; pick up another pin; remove practice pins		d4400 - Picking up; d4402 - Manipulating; d4402 - Manipulating; d4400 - Picking up; d4403 - Releasing;		

	starting with the top hole. Work as rapidly as you can until I say 'Stop.' Are you ready? Begin." "Stop"								
	Both hands								
	"For this part of the test, you will use both hands at the same time. Pick up a pin from the right-handed cup with your right hand, and at the same time pick up a pin from the left-handed cup with your left hand. Then place the pins down the rows. Begin with the top hole of both rows. (Demonstrate. Then replace the pins used for demonstration.) Now you may insert a few pins with both hands for practice." "Stop. Take out the practice pins, and put them back in their cups." "When I say 'Begin,' place as many pins as possible with both hands, starting with the top hole	Capacity	N/A	N/A	pick up; place each pin; insert a few pins; pick up another pin; remove practice pins		d4400 - Picking up; d4402 - Manipulating; d4402 - Manipulating; d4400 - Picking up; d4403 - Releasing;		

	of both rows. Work as rapidly as you can, until I say 'Stop.' Are you ready? Begin." "Stop"								
	Right + Left + Both hands Assembly								
	"Pick up one pin from the right-handed cup with your right hand. While you are placing it in the top hole in the right-handed row, pick up a washer with your left hand. As soon as the pin has been placed, drop the washer over the pin. While the washer is being placed over the pin with you left hand, pick up a collar with your right hand. While the collar is being dropped over the pin, pick up another washer with your left hand and drop it over the collar. This completes the first 'assembly,'	Capacity	N/A	N/A	pick up one pin; placing it in top; pick up washer; drop washer; pick up collar; drop pin	coordination	d4400 - Picking up; d4402 - Manipulating; d4400 - Picking up; d4403 - Releasing; d4400 - Picking up; d4403 - Releasing	b7602 - Coordination of voluntary movements	

	consisting of a pin, a washer, a collar, and a washer. While the final washer for the first assembly is being placed with your left hand, start the second assembly immediately by picking up another pin with your right hand. Place it in the next hole; drop a washer over it with your left hand, and so on, completing another assembly. Now take a moment to try a few practice assemblies." "Sto p. Now return the pins, collars, and washers to their proper cups." "When I say 'Begin,' make as many assemblies as possible, beginning with the top hole. Work quickly until I say 'Stop.'								
Motor Function Measure (MFM)	*Supine*								
	Raises one hand from the mat and moves it to the opposite shoulder	Capacity	N/A	N/A	raise hand and move to opposite shoulder		b7602 - Coordination of voluntary movements		

	Seated on the chair or in their wheelchair								
	Forearms on the table but not elbows: raises both hands to the top of the head at the same time, head and trunk in the axis	Capacity	N/A	N/A	raises both head to top of head at the same time		b7602 - Coordination of voluntary movements		
	The pencil on the table: reaches the pencil with one hand, elbow in complete extension at the end of the movement	Capacity	N/A	N/A	reaches	elbow in complete ext	d4452 - Reaching	b7100 - Mobility of a single joint	
	10 coins placed on the table: successively picks up and holds 10 coins in one hand within 20 s	Capacity	N/A	N/A	picks up coins	holds coins	d4400 - Picking up	d4401 - Grasping	
	One finger placed in the center of the fixed CD: traces the complete border of the disk with one finger without support of the hand	Capacity	N/A	N/A	traces		b7602 - Coordination of voluntary movements		
	The pencil on the table: picks up the pencil placed next to their hand and draws a continuous series of loops of 1 cm	Capacity	N/A	N/A	picks up pencil	draws	d4400 - Picking up	d4408 - Fine hand use, other specified	drawing

	height in the 4-cm-long frame								
	Holding the sheet of paper: tears the paper folded in 4, beginning at the fold	Capacity	N/A	N/A	tear paper		d4402 - Manipulating		
	The tennis ball on the table: picks up the ball, raises it off the table and turns over the hand holding onto the ball	Capacity	N/A	N/A	pick up ball	raising and turning over the hand	d4400 - Picking up	b7602 - Coordination of voluntary movements	
	A finger placed in the center of the fixed square: raises the finger and places it successively in the center of the 8 squares of the diagram without touching the lines	Capacity	N/A	N/A	raises finger and places it successfully in center		b7602 - Coordination of voluntary movements		
	Upper limbs along the trunk: places the two forearms and/or hands on the table at the same time	Capacity	N/A	N/A	places two forearms at same time		b7602 - Coordination of voluntary movements		
Functional Dexterity Test (FDT)	*Non-injured hand first*								
	"Please start with your non-injured hand. Start by turning the peg at the top opposite corner [point to peg], turn all the pegs over as	Capacity	N/A	N/A	turning the peg; supinate; drop peg; pick up peg; put it in pegboard;	coordination	d4453 - Turning or twisting the hands or arms; b7600 - Control of simple voluntary movements; d4403 -	b7602 - Coordination of voluntary movements	

	quickly as possible, turning over one row of pegs, then reversing the order in the next row, in a zigzag fashion" “Do not turn your hand up to face the ceiling (supinate) or touch the board for help in turning the peg; each of these motions carries a penalty of 5 seconds. If you drop a peg, time is stopped, and a 10-second penalty is added. You then need to retrieve the peg and put it in the pegboard in the unturned position. Then continue to turn the pegs with the peg that you just put back. The clock starts where it was stopped, and the time is continued.” “Turn over all the pegs on the board with your non-injured hand.”						Releasing; d4400 - Picking up; d4402 - Manipulating		
	Injured hand								
	Same as above	N/A	N/A	N/A					
Hand Assessment	B Grasps from an easy position	Capacity	N/A	N/A	grasps		d4401 - Grasping		

of Infants (HAI)	B Holds for a while	Capacity	N/A	N/A	hold		d4401 - Grasping		
	B Moves fingers	Capacity	N/A	N/A	move fingers		b7600 - Control of simple voluntary movements		
	B Quality of movements	Capacity	N/A	N/A	quality of movement		b7101 - Mobility of several joints		
	B Quality of holding	Capacity	N/A	N/A	quality of holding		d4401 - Grasping		
	B Object location when grasping	Capacity	N/A	N/A	object location when grasping		b7602 - Coordination of voluntary movements		
	B Amount of hand use	Capacity	N/A	N/A	hand use		d4409 - Fine hand use, unspecified		general hand use
	B Initiates use for objects in midline	Capacity	N/A	N/A	initiates use		d2100 - Undertaking a simple task		
	B Initiates use for objects on the side	Capacity	N/A	N/A	initiates use		d2100 - Undertaking a simple task		
	B Moves upper arm	Capacity	N/A	N/A	moves upper arm		b7101 - Mobility of several joints		
	B Moves forearm	Capacity	N/A	N/A	moves forearm		b7100 - Mobility of a single joint		
	B Adjusts arm/hand orientation	Capacity	N/A	N/A	adjust hand/arm		b7600 - Control of simple voluntary movements		
	L Moves upper arm	Capacity	N/A	N/A	moves upper arm		b7101 - Mobility of several joints		
	L Holds for a while	Capacity	N/A	N/A	hold		d4401 - Grasping		
	L Moves fingers	Capacity	N/A	N/A	moves finger		b7101 - Mobility of several joints		
	L Grasps from an easy position	Capacity	N/A	N/A	grasps		d4401 - Grasping		

	Bim Bilateral holding of object	Capacity	N/A	N/A	grasps		d4401 - Grasping		
	L Moves forearm	Capacity	N/A	N/A	moves forearm		b7100 - Mobility of a single joint		
	L Initiates use for objects on the side	Capacity	N/A	N/A	initiates use		d2100 - Undertaking a simple task		
	Bim Bilateral object contact	Capacity	N/A	N/A	touch		d1201 - Touching		
	L Quality of holding	Capacity	N/A	N/A	quality of holding		d4401 - Grasping		
	L Object location when grasping	Capacity	N/A	N/A	object location		b7602 - Coordination of voluntary movements		
	Bim Transfers between hands	Capacity	N/A	N/A	transfers between hands		b7602 - Coordination of voluntary movements		
	L Initiates use for objects in midline	Capacity	N/A	N/A	initiates use		b7100 - Mobility of a single joint		
	Bim Bilateral manipulation	Capacity	N/A	N/A	manipulation		d4402 - Manipulating		
	L Adjusts arm/hand orientation	Capacity	N/A	N/A	adjust arm/hand		b7600 - Control of simple voluntary movements		
	L Amount of hand use	Capacity	N/A	N/A	hand use		d4409 - Fine hand use, unspecified		general hand use
	L Quality of movements	Capacity	N/A	N/A	quality of movement		b7101 - Mobility of several joints		
	Bim Transfers in a sequence	Capacity	N/A	N/A	hand transfers		b7602 - Coordination of voluntary movements		
Pediatric Arm	*Unilateral items*								
	Reach above head	Capacity	N/A	N/A	reaching		d4452 - Reaching		

Function Test (PAFT)	Reach at waist level	Capacity	N/A	N/A	reaching		d4452 - Reaching		
	Reach across midline	Capacity	N/A	N/A	reaching		d4452 - Reaching		
	Grasp ball	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Carry ball	Capacity	N/A	N/A	carry		d4301 - Carrying in the hands		
	Release ball into cup	Capacity	N/A	N/A	release		d4403 - Releasing		
	Pour ball out of cup	Capacity	N/A	N/A	pouring		d4453 - Turning or twisting the hands or arms		
	Throw ball onto target	Capacity	N/A	N/A	throw		d4454 - Throwing		
	Isolated finger use	Capacity	N/A	N/A	finger use		b7600 - Control of simple voluntary movements		
	Removing big-knob puzzle piece	Capacity	N/A	N/A	removing puzzle		d4402 - Manipulating		
	Crayon grasp	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Crayon use	Capacity	N/A	N/A	crayon use		d4409 - Fine hand use, unspecified		general use
	Grasp cracker-sized food (e.g. animal or graham cracker, saltine)	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Grasp small food item (e.g. cheerio or raisin)	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Eat with a spoon	Capacity	N/A	N/A	eating		d550 - Eating		
	Bilateral items								
	Separate pull-apart toy	Capacity	N/A	N/A	pull apart		d4458 - Hand and arm use, other specified		pull apart

	Carry large ball (e.g. basketball)	Capacity	N/A	N/A	carry		d4302 - Carrying in the arms		
	Throw ball into hoop	Capacity	N/A	N/A	throw		d4454 - Throwing		
	Place hat on head	Capacity	N/A	N/A	hat on head		d5400 - Putting on clothes		
	Put on boots (using hands)	Capacity	N/A	N/A	put on boots		d5402 - Putting on footwear		
	Quadruped weight-bearing	Capacity	N/A	N/A	weight bearing four limbs		b7603 - Supportive functions of arm or leg		
	Crawling	Capacity	N/A	N/A	crawling		d4550 - Crawling		
Upper Limb Physician Rating Scale (ULPRS)	Active elbow extension (normal 180 degrees)	Capacity	N/A	N/A	elbow ext		b7100 - Mobility of a single joint		
	Active supination in extension (Elbow extended, forearm supinates) mid-position: palm to 90 degree horizontal	Capacity	N/A	N/A	elbow ext + supination		b7101 - Mobility of several joints		
	Active supination in flexion (elbow flexed at 90 degree forearm supinates)	Capacity	N/A	N/A	supination in flexion		b7101 - Mobility of several joints		
	Active wrist dorsiflexion (forearm supported, active dorsiflexion of wrist) mid-position: palm level with forearm	Capacity	N/A	N/A	wrist dorsiflexion		b7100 - Mobility of a single joint		
	Wrist dorsiflexion (angle of movement)	Capacity	N/A	N/A	wrist dorsiflexion		b7100 - Mobility of a single joint		

	Finger opening	Capacity	N/A	N/A	finger opening		b7101 - Mobility of several joints		
	Thumb in function	Capacity	N/A	N/A	thumb movement		b7100 - Mobility of a single joint		
	Associated increase in muscle tone	Capacity	N/A	N/A	increased muscle tone		b7350 - Tone of isolated muscles and muscle groups		
	Two handed function	Capacity	N/A	N/A	bilateral function		b7602 - Coordination of voluntary movements		
Assessment of Children's Hand Skills	Manual gesture	Capacity	N/A	N/A	gesture		d4409 - Fine hand use, unspecified		general manual gesture
	Body contact hand skills	Capacity	N/A	N/A	hand skills		d4409 - Fine hand use, unspecified		hand skills
	Grasping	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Holding	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	In-hand manipulation	Capacity	N/A	N/A	manipulation		d4402 - Manipulating		
	Releasing	Capacity	N/A	N/A	release		d4403 - Releasing		
	Isolated finger movement	Capacity	N/A	N/A	finger movement		b7600 - Control of simple voluntary movements		
	Reaching	Capacity	N/A	N/A	reaching		d4452 - Reaching		
	Turning	Capacity	N/A	N/A	turning		d4453 - Turning or twisting the hands or arms		
	Carrying	Capacity	N/A	N/A	carrying		d4309 - Lifting and carrying, unspecified		general carrying
	Throwing	Capacity	N/A	N/A	throwing		d4454 - Throwing		

	Catching	Capacity	N/A	N/A	catching		d4455 - Catching		
	Moving	Capacity	N/A	N/A	moving		d4459 - Hand and arm use, unspecified		general moving
	Stabilizing	Capacity	N/A	N/A	stabilizing		b7159 - Stability of joint functions, unspecified		general stabilization
	Transferring	Capacity	N/A	N/A	transferring		d4409 - Fine hand use, unspecified		general transferring
	Using both hands simultaneously	Capacity	N/A	N/A	simultaneous movement		b7602 - Coordination of voluntary movements		
	Using both hands cooperatively	Capacity	N/A	N/A	coordination		b7602 - Coordination of voluntary movements		
	Accuracy	Capacity	N/A	N/A	control		b7601 - Control of complex voluntary movements		
	Pace	Capacity	N/A	N/A	pace		b7609 - Control of voluntary movement functions, unspecified		pace
	Movement quality	Capacity	N/A	N/A	movement quality		b7101 - Mobility of several joints		
Test of Gross Motor Development -2	1. Striking a stationary ball (the ability to strike a stationary ball with a plastic bat)	Capacity	N/A	N/A	striking		d4458 - Hand and arm use, other specified		striking
	2. Stationary dribble (the ability to dribble a basket ball a	Capacity	N/A	N/A	dribble		d4458 - Hand and arm use, other specified		dribbling ball

	minimum of four times with the dominant hand before catching the ball with both hands, without moving feet)								
	3. Catch (the ability to catch a plastic ball that has been tossed underhand)	Capacity	N/A	N/A	catching		d4455 - Catching		
	4. Overhand throw (the ability to throw a ball at a point on a wall with the preferred hand)	Capacity	N/A	N/A	throwing		d4454 - Throwing		
	5. Underhand roll (the ability to roll a ball between two cones with the preferred hand)	Capacity	N/A	N/A	rolling		d4458 - Hand and arm use, other specified		rolling
Besta Scale	*Grip (three cubes of different sizes)*	Capacity	N/A	N/A					
	Pick up the cubes first with the unaffected hand and then with the impaired hand	Capacity	N/A	N/A	pick up		d4400 - Picking up		
	Spontaneous use of affected hand during bilateral manipulation	Capacity	N/A	N/A					
	Throw a large ball	Capacity	N/A	N/A	throw		d4454 - Throwing		
	Tear a sheet of paper into many pieces	Capacity	N/A	N/A	tear sheet		d4402 - Manipulating		

	Unscrew and screw the cap of a bottle	Capacity	N/A	N/A	unscrew	screw	d4453 - Turning or twisting the hands or arms	d4453 - Turning or twisting the hands or arms	
	Open a packet tied with adhesive tape	Capacity	N/A	N/A	open packet		d4408 - Fine hand use, other specified		opening
	Open a packet tied with string in a single knot	Capacity	N/A	N/A	open packet		d4408 - Fine hand use, other specified		opening
	Wrap an object in paper forming a parcel	Capacity	N/A	N/A	wrap an object		d4408 - Fine hand use, other specified		wrapping
	Cut out geometrical figures and stick them onto a sheet of paper	Capacity	N/A	N/A	cut out	sticking	d4402 - Manipulating	d4402 - Manipulating	
	Fold a piece of paper and place it in an envelope	Capacity	N/A	N/A	folding paper	place it envelop	d4402 - Manipulating	d4402 - Manipulating	
	Stereognosis								
	Small spoon	Capacity	N/A	N/A	sensation of touch		b265 - Touch function		
	Coin	Capacity	N/A	N/A	sensation of touch		b265 - Touch function		
	Brush	Capacity	N/A	N/A	sensation of touch		b265 - Touch function		
	Small ball	Capacity	N/A	N/A	sensation of touch		b265 - Touch function		
	Doll	Capacity	N/A	N/A	sensation of touch		b265 - Touch function		
Selective Control of the Upper Extremity Scale (SCUES)	Selective motor control - shoulder	Capacity	N/A	N/A	Selective motor control - shoulder		b7601 - Control of complex voluntary movements		
	Selective motor control - elbow	Capacity	N/A	N/A	Selective motor control - elbow		b7601 - Control of complex		

							voluntary movements		
	Selective motor control - forearm	Capacity	N/A	N/A	Selective motor control - forearm		b7601 - Control of complex voluntary movements		
	Selective motor control - wrist	Capacity	N/A	N/A	Selective motor control - wrist		b7601 - Control of complex voluntary movements		
	Selective motor control - fingers/thumb	Capacity	N/A	N/A	Selective motor control - fingers/thumb		b7601 - Control of complex voluntary movements		
Bayley Scales of Infant and Toddler Development (BSID) - 3	*Fine Motor subtest*								
	Hands are fisted	Capacity	N/A	N/A	hands fisted		b7350 - Tone of isolated muscles and muscle groups		
	Attempts to bring hand to mouth	Capacity	N/A	N/A	bring hand to mouth		b7602 - Coordination of voluntary movements		
	Retains ring [ring with string]	Capacity	N/A	N/A	hold		d4401 - Grasping		
	Keeps hands open	Capacity	N/A	N/A	hand open		b7350 - Tone of isolated muscles and muscle groups		
	Rotates wrist [ring with string]	Capacity	N/A	N/A	rotate wrist		b7100 - Mobility of a single joint		
	Grasps suspended ring [ring with string]	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Reaches for block [block with hole]	Capacity	N/A	N/A	reaching		d4452 - Reaching		
	Touches block [block with hole]	Capacity	N/A	N/A	touch		b7602 - Coordination of voluntary movements		

	Whole hand grasp [block with hole]	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Reaches unilaterally	Capacity	N/A	N/A	reaching		d4452 - Reaching		
	Raking grasp [food pellet]	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Partial thumb opposition [block with hole]	Capacity	N/A	N/A	thumb opposition		b7100 - Mobility of a single joint		
	Transfers ring [ring]	Capacity	N/A	N/A	transferring hand		b7602 - Coordination of voluntary movements		
	Whole hand grasp [food pellet]	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Transfers block [block without hole]	Capacity	N/A	N/A	transferring block		b7602 - Coordination of voluntary movements		
	Thumb-fingertip grasp [block without hole]	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Blocks to midline [2 blocks without hole]	Capacity	N/A	N/A	midline		b7602 - Coordination of voluntary movements		
	Partial thumb opposition [food pellet]	Capacity	N/A	N/A	thumb opposition		b7100 - Mobility of a single joint		
	Lifts cup by handle [cup with handle]	Capacity	N/A	N/A	lift cup		d4300 - Lifting		
	Thumb finger-tip grasp [food pellet]	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	turns pages of book [picture book]	Capacity	N/A	N/A	turn page		d4402 - Manipulating		
	palmar grasp [crayon or pencil]	Capacity	N/A	N/A	grasp		d4401 - Grasping		

	isolates extended index finger [pegboard]	Capacity	N/A	N/A	extend finger		b7100 - Mobility of a single joint		
	scribbles spontaneously [crayon or pencil]	Capacity	N/A	N/A	scribble		d1450 - Acquiring skills to use writing implements		
	block stacking 2 blocks [blocks]	Capacity	N/A	N/A	stacking blocks		d4402 - Manipulating		stacking
	imitates random stroke [2 crayons]	Capacity	N/A	N/A	imitates random stroke		d1450 - Acquiring skills to use writing implements		
	places 10 pellets in bottle [food pellets]	Capacity	N/A	N/A	places pellets in bottle		d4402 - Manipulating		
	transitional grasp [crayon or pencil]	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	coins in slot [bank and pennies]	Capacity	N/A	N/A	coins in slot		d4402 - Manipulating		
	connecting blocks: apart	Capacity	N/A	N/A	pull apart blocks		d4458 - Hand and arm use, other specified		pull apart
	intermediate tripod grasp [crayon or pencil]	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	block stacking 6 blocks [blocks]	Capacity	N/A	N/A	stacking blocks		d4402 - Manipulating		
	uses hand to hold paper in place [crayon or pencil and paper]	Capacity	N/A	N/A	hand to stabilize		b7603 - Supportive functions of arm or leg		
	imitates horizontal stroke [crayon or pencil and paper]	Capacity	N/A	N/A	imitates horizontal stroke		d1450 - Acquiring skills to use writing implements		
	imitates vertical stroke [crayon or pencil and paper]	Capacity	N/A	N/A	imitates vertical stroke		d1450 - Acquiring skills to use writing implements		

	connecting blocks: together	Capacity	N/A	N/A	connecting blocks		d4402 - Manipulating		
	imitates circular stroke [crayon or pencil and paper]	Capacity	N/A	N/A	imitates circular stroke		d1450 - Acquiring skills to use writing implements		
	builds train of blocks [10 blocks]	Capacity	N/A	N/A	builds train		d4402 - Manipulating		
	String 3 blocks [shoelace and blocks]	Capacity	N/A	N/A	stringing 3 blocks		d4402 - Manipulating		
	imitates hand movement	Capacity	N/A	N/A	imitates hand movement		b7602 - Coordination of voluntary movements		
	snips paper [paper and scissors]	Capacity	N/A	N/A	snips paper		d4402 - Manipulating		
	grasp dynamic grasp [crayon or pencil]	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	Tactilely discriminates shapes [pegs, blocks without hole, square pieces, drawstring bag]	Capacity	N/A	N/A	tactile discrimination		b265 - Touch function		
	Builds wall [blocks]	Capacity	N/A	N/A	builds wall		d4402 - Manipulating		
	Cuts paper [paper and scissors]	Capacity	N/A	N/A	cuts paper		d4402 - Manipulating		
	Builds bridge [blocks]	Capacity	N/A	N/A	builds bridge		d4402 - Manipulating		
	imitates plus sign [crayon or pencil and paper]	Capacity	N/A	N/A	imitates plus sign		d1450 - Acquiring skills to use writing implements		
	block stacking 8 blocks [blocks]	Capacity	N/A	N/A	block stacking		d4402 - Manipulating		

	cuts on line [crayon or pencil and paper]	Capacity	N/A	N/A	cuts on line		d4402 - Manipulating		
	Builds T [blocks]	Capacity	N/A	N/A	builds T		d4402 - Manipulating		
	buttons 1 button [button sleeve]	Capacity	N/A	N/A	buttons 1 button		d4408 - Fine hand use, other specified		buttoning
	builds steps [blocks]	Capacity	N/A	N/A	builds steps		d4402 - Manipulating		
	traces designs [pencil traces sheet]	Capacity	N/A	N/A	traces		b7602 - Coordination of voluntary movements		
	imitates square [crayon or pencil and paper]	Capacity	N/A	N/A	imitates square		d1450 - Acquiring skills to use writing implements		
	copies plus sign [crayon or pencil and paper]	Capacity	N/A	N/A	copies plus sign		d1450 - Acquiring skills to use writing implements		
	taps finger	Capacity	N/A	N/A	taps finger		b7100 - Mobility of a single joint		
	places 20 pellets in bottle [food pellets]	Capacity	N/A	N/A	places pellets in bottle		d4402 - Manipulating		
	cuts circle [paper and scissors]	Capacity	N/A	N/A	cuts circle		d4402 - Manipulating		
	cuts square [paper and scissors]	Capacity	N/A	N/A	cuts square		d4402 - Manipulating		
	copies square [crayon or pencil and paper]	Capacity	N/A	N/A	copies square		d1450 - Acquiring skills to use writing implements		
Minnesota Handwriting Assessment	Child asked to write out the sentence	Performance	N/A	N/A	write out sentence		d1701 - Using grammatical conventions in writing compositions		

Mini Assisting Hands Assessment (Mini-AHA)	Bimanual manipulation	Capacity	N/A	N/A	manipulation		d4402 - Manipulating		
	chooses assisting hand	Capacity	N/A	N/A	assisting hand		d4459 - Hand and arm use, unspecified		general assisting hand
	varies grasp	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	grasps (object location)	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	adjusts arm position	Capacity	N/A	N/A	arm position		b7602 - Coordination of voluntary movements		
	symmetrical arm/hand use	Capacity	N/A	N/A	symmetrical arm use		b7602 - Coordination of voluntary movements		
	readjust grasp	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	flow in bimanual performance	Capacity	N/A	N/A	bimanual		b7602 - Coordination of voluntary movements		
	releases	Capacity	N/A	N/A	releases		d4403 - Releasing		
	initiates use	Capacity	N/A	N/A	initiates use		d2100 - Undertaking a simple task		
	reaches	Capacity	N/A	N/A	reaches		d4452 - Reaching		
	stabilize by grasp	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	moves forearm	Capacity	N/A	N/A	moves forearm		b7100 - Mobility of a single joint		
	moves fingers	Capacity	N/A	N/A	moves fingers		b7101 - Mobility of several joints		
	moves upper arm	Capacity	N/A	N/A	moves upper arm		b7100 - Mobility of a single joint		

	amount of use	Capacity	N/A	N/A	amount of use		d4409 - Fine hand use, unspecified		general use
	stabilize by weight	Capacity	N/A	N/A	stabilize		b7603 - Supportive functions of arm or leg		
	grasp from an easy position	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	contact with objects	Capacity	N/A	N/A	contact with object		d1201 - Touching		
	holds	Capacity	N/A	N/A	hold		d4401 - Grasping		
Both Hands Assessment (BoHa)	Manipulates	Capacity	N/A	N/A	manipulation		d4402 - Manipulating		
	grip force regulation	Capacity	N/A	N/A	grasp	force regulation	d4401 - Grasping	b7301 - Power of muscles of one limb	
	proceeds	Capacity	N/A	N/A	proceeds		d4409 - Fine hand use, unspecified		general fine hand use
	flow in bimanual performance	Capacity	N/A	N/A	flow in bimanual		b7602 - Coordination of voluntary movements		
	coordinates	Capacity	N/A	N/A	coordinates		b7602 - Coordination of voluntary movements		
	stabilize objects	Capacity	N/A	N/A	stabilize object		b7603 - Supportive functions of arm or leg		
	moves fingers	Capacity	N/A	N/A	moves finger		b7101 - Mobility of several joints		
	varies type of grasp	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	readjust grasp	Capacity	N/A	N/A	grasp		d4401 - Grasping		

	orients object	Capacity	N/A	N/A	orients objects		d4402 - Manipulating		
	quality of arm movements	Capacity	N/A	N/A	quality of arm movement		b7101 - Mobility of several joints		
	grasps	Capacity	N/A	N/A	grasp		d4401 - Grasping		
	speed of movements	Capacity	N/A	N/A	speed of movement		b7602 - Coordination of voluntary movements		
	releases	Capacity	N/A	N/A	releases		d4403 - Releasing		
	reaches	Capacity	N/A	N/A	reaches		d4452 - Reaching		
	initiates	Capacity	N/A	N/A	initiates		d2100 - Undertaking a simple task		
Unilateral Below Elbow Test (UBET)	*2-4 year old*								
	take play doh out of a plastic bag	Capacity	N/A	N/A	take out		d4400 - Picking up		
	bang cymbals together	Capacity	N/A	N/A	bang together		d4458 - Hand and arm use, other specified		bang together
	put sock on foot	Capacity	N/A	N/A	put on socks		d5402 - Putting on footwear		
	thread beads	Capacity	N/A	N/A	threading		d4402 - Manipulating		
	open a jar of bubbles	Capacity	N/A	N/A	open jar		d4453 - Turning or twisting the hands or arms		
	ride on a rolling racer	Capacity	N/A	N/A	ride on rolling racer		d4153 - Maintaining a sitting position		
	open drawstring bag and dump lego duplo out	Capacity	N/A	N/A	open drawstring	dump out	d4402 - Manipulating	d4402 - Manipulating	
	Separate Lego Duplo	Capacity	N/A	N/A	pull apart		d4458 - Hand and arm use, other specified		pull apart

	open a box of crayons and remove one	Capacity	N/A	N/A	open box	remove crayon	d4408 - Fine hand use, other specified	d4400 - Picking up	open box
	5-7 year old								
	cut paper circle from construction paper	Capacity	N/A	N/A	cutting paper		d4402 - Manipulating		
	remove cap from felt tip marker	Capacity	N/A	N/A	remove cap from marker		d4408 - Fine hand use, other specified		remove cap
	sharpen pencil	Capacity	N/A	N/A	sharpen pencil		d4408 - Fine hand use, other specified		sharpen pencil
	do up buttons on vest	Capacity	N/A	N/A	buttoning		d4408 - Fine hand use, other specified		buttoning
	tie shoelaces in a knot	Capacity	N/A	N/A	ties shoe laces		d4402 - Manipulating		
	turn kaleidoscope	Capacity	N/A	N/A	turning		d4453 - Turning or twisting the hands or arms		
	separate legos	Capacity	N/A	N/A	separate legos		d4408 - Fine hand use, other specified		separate legos
	use bow and arrow	Capacity	N/A	N/A	use bow and arrow	sport	d4458 - Hand and arm use, other specified	d9201 - Sports	use bow and arrow
	ride on rolling racer	Capacity	N/A	N/A	ride on rolling racer		d4153 - Maintaining a sitting position		
	8-10 year old								
	swing a bat	Capacity	N/A	N/A	swinging bat	sport	d4458 - Hand and arm use, other specified	d9201 - Sports	swinging bat
	winds string onto yo-yo	Capacity	N/A	N/A	winding string		d4458 - Hand and arm use, other specified		winding string
	open a band aid	Capacity	N/A	N/A	open band aid		d4402 - Manipulating		

	tie shoelaces in a bow	Capacity	N/A	N/A	tie shoelace		d4402 - Manipulating		
	do up buttons on a shirt	Capacity	N/A	N/A	buttoning		d4408 - Fine hand use, other specified		buttoning
	make a telescope with paper and a rubber band	Capacity	N/A	N/A	manipulation	crafts	d4402 - Manipulating	d9203 - Crafts	
	place glove on unaffected hand	Capacity	N/A	N/A	dressing		d5400 - Putting on clothes		
	draw a line with a ruler	Capacity	N/A	N/A	drawing		d4408 - Fine hand use, other specified		drawing
	start zipper on vest	Capacity	N/A	N/A	zipper		d4408 - Fine hand use, other specified		zipper
	11-21 year old								
	cut paper from a roll to wrap a videotape	Capacity	N/A	N/A	cut paper		d4402 - Manipulating		
	tear a piece of tape	Capacity	N/A	N/A	tearing		d4402 - Manipulating		
	secure wrap no a videotape with tape	Capacity	N/A	N/A	secure wrap		d4408 - Fine hand use, other specified		secure wrap
	cut putty on plate with knife and fork	Capacity	N/A	N/A	cutting		d4402 - Manipulating		
	open a three ring binder	Capacity	N/A	N/A	open binder		d4408 - Fine hand use, other specified		open binder
	start zipper on vest	Capacity	N/A	N/A	zipper		d4408 - Fine hand use, other specified		zipper
	tie shoelaces in a bow	Capacity	N/A	N/A	tie shoe laces		d4402 - Manipulating		
	do up buttons on a shirt	Capacity	N/A	N/A	buttoning		d4408 - Fine hand use, other specified		buttoning

	use dust pan and small broom	Capacity	N/A	N/A	dust pan and broom		d6408 - Doing housework, other specified		dust pan and broom
Brachial Plexus outcome measure (BPOM)	combs back of head	Capacity	N/A	N/A	combing		d5202 - Caring for hair		
	pulls apart theraputty	Capacity	N/A	N/A	pull apart		d4458 - Hand and arm use, other specified		
	opens large container	Capacity	N/A	N/A	opening		d4408 - Fine hand use, other specified		
	holds plate with palm up	Capacity	N/A	N/A	hold	palm up	d4401 - Grasping	b7100 - Mobility of a single joint	
	pretends to eat candy	Capacity	N/A	N/A	eat		d550 - Eating		
	strings bead	Capacity	N/A	N/A	stringing bead		d4402 - Manipulating		
	plays drums	Capacity	N/A	N/A	playing drums		d9202 - Arts and culture		
	places container above head	Capacity	N/A	N/A	lifting above head		d4300 - Lifting		
	uses computer mouse	Capacity	N/A	N/A	uses computer mouse		d4402 - Manipulating		
	undo button at midline	Capacity	N/A	N/A	unbutton		d4408 - Fine hand use, other specified		
	hand to back pant pocket	Capacity	N/A	N/A	hand to back pant pocket		b7602 - Coordination of voluntary movements		
In Hand Manipulation Test	picking up coins and placing them in a bank	Capacity	N/A	N/A	picking up coins	placing them	d4400 - Picking up	d4402 - Manipulating	
	picking up chips and placing them in a container	Capacity	N/A	N/A	pick up chips	placing them	d4400 - Picking up	d4402 - Manipulating	
	removing and replacing small bottle lids	Capacity	N/A	N/A	removing lid		d4453 - Turning or twisting the hands or arms		

	picking up and placing cubes; turning over cubes	Capacity	N/A	N/A	picking up coins	placing; turning over	d4400 - Picking up	d4402 - Manipulating	
	picking up, turning over, and placing pegs	Capacity	N/A	N/A	pick up	turning and placing	d4400 - Picking up	d4402 - Manipulating	
	picking writing tools	Capacity	N/A	N/A	picking writing tools		d4400 - Picking up		
	putting a key into a lock	Capacity	N/A	N/A	putting key into lock		d4402 - Manipulating		
	turning pages in a magazine	Capacity	N/A	N/A	turning pages		d4402 - Manipulating		
	picking up playing cards	Capacity	N/A	N/A	picking up playing cards		d4400 - Picking up		
Assessment of Capacity for Myoelectric Control (ACMC)	*Gripping*								
	grips with weight supported	Capacity	N/A	N/A	grips		d4401 - Grasping		
	positions the hand and grips	Capacity	N/A	N/A	grips	position hands	d4401 - Grasping	b7602 - Coordination of voluntary movements	
	uses the tripod pinch grip with weight supported	Capacity	N/A	N/A	grips		d4401 - Grasping		
	positions the hand and uses the tripod pinch grip	Capacity	N/A	N/A	grips	position hands	d4401 - Grasping	b7602 - Coordination of voluntary movements	
	adjusts grip force to avoid crushing	Capacity	N/A	N/A	grips	avoid crushing	d4401 - Grasping	b7300 - Power of isolated muscles and muscle groups	
	grips with the arm in different positions	Capacity	N/A	N/A	grips	arm different position	d4401 - Grasping	b7101 - Mobility of several joints	

	grips through iterative refinement, manipulates	Capacity	N/A	N/A	grips	manipulation	d4401 - Grasping	d4402 - Manipulating	
	grips object by feeding arm forwards	Capacity	N/A	N/A	reach		d4452 - Reaching		
	grips object moving towards the hand	Capacity	N/A	N/A	catch		d4455 - Catching		
	grips with no visual feedback	Capacity	N/A	N/A	grips		d4401 - Grasping		
	adjusts grip force with no visual feedback	Capacity	N/A	N/A	grip force		b7300 - Power of isolated muscles and muscle groups		
	grips behind the back to manipulate object	Capacity	N/A	N/A	grips	manipulate	d4401 - Grasping	d4402 - Manipulating	
	Holding								
	holds with arm supported	Capacity	N/A	N/A	hold	arm supported	d4401 - Grasping	b7603 - Supportive functions of arm or leg	
	holds without support	Capacity	N/A	N/A	hold		d4401 - Grasping		
	holds without crushing	Capacity	N/A	N/A	hold	without crushing	d4401 - Grasping	b7300 - Power of isolated muscles and muscle groups	
	holds with the arm moving	Capacity	N/A	N/A	hold	arm moving	d4401 - Grasping	b7101 - Mobility of several joints	
	holds with no visual feedback	Capacity	N/A	N/A	hold		d4401 - Grasping		
	holds with the arm moving, no visual feedback	Capacity	N/A	N/A	hold	arm moving	d4401 - Grasping	b7101 - Mobility of	

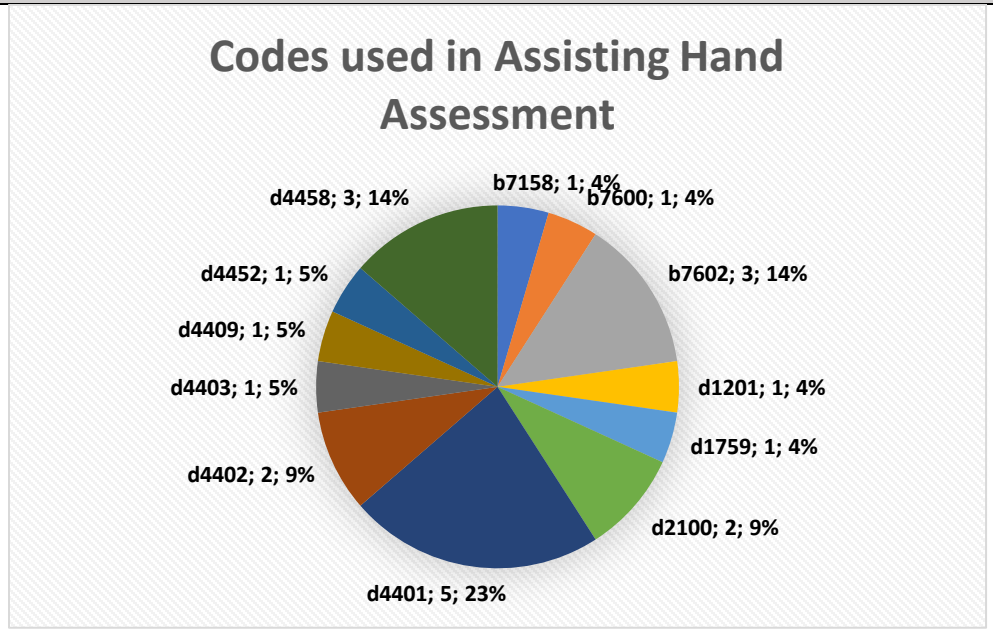
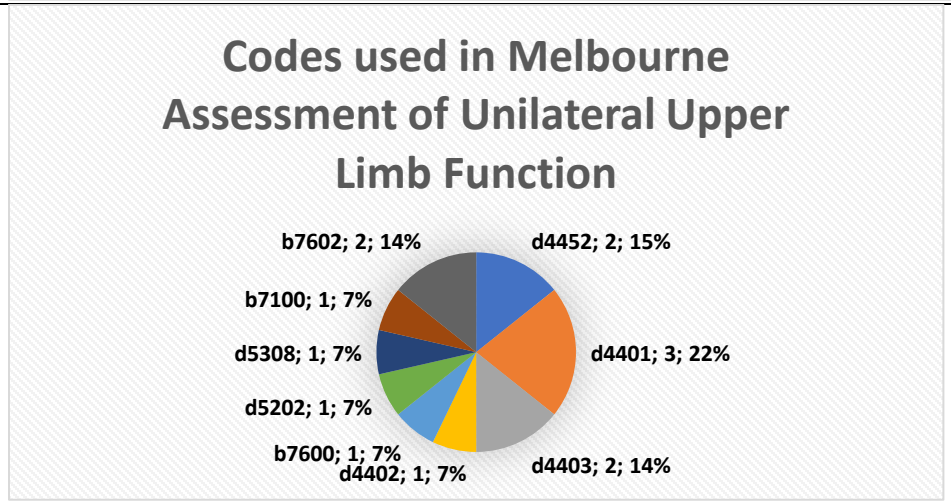
								several joints	
	Releasing								
	releases with arm supported	Capacity	N/A	N/A	release	arm supported	d4403 - Releasing	b7603 - Supportive functions of arm or leg	
	positions the hand and releases	Capacity	N/A	N/A	release	position hand	d4403 - Releasing	b7101 - Mobility of several joints	
	loosens grip without dropping object	Capacity	N/A	N/A	release	without dropping	d4403 - Releasing	b7300 - Power of isolated muscles and muscle groups	
	releases the grip with arm in different positions	Capacity	N/A	N/A	release	different position	d4403 - Releasing	b7602 - Coordination of voluntary movements	
	releases through iterative refinement, manipulation	Capacity	N/A	N/A	release	manipulation	d4403 - Releasing	d4402 - Manipulating	
	releases object with arm swinging low	Capacity	N/A	N/A	release		d4403 - Releasing		
	releases object by feeding arm forward at, or above, shoulder height	Capacity	N/A	N/A	release	reaching	d4403 - Releasing	d4452 - Reaching	
	releases synchronized with the other hand	Capacity	N/A	N/A	release	synchronize with other hand	d4403 - Releasing	b7602 - Coordination of voluntary movements	
	releases with no visual feedback	Capacity	N/A	N/A	release		d4403 - Releasing		

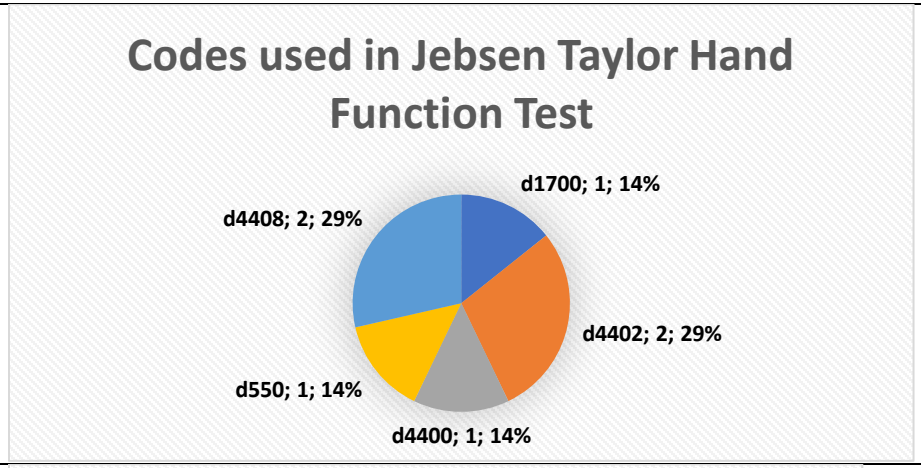
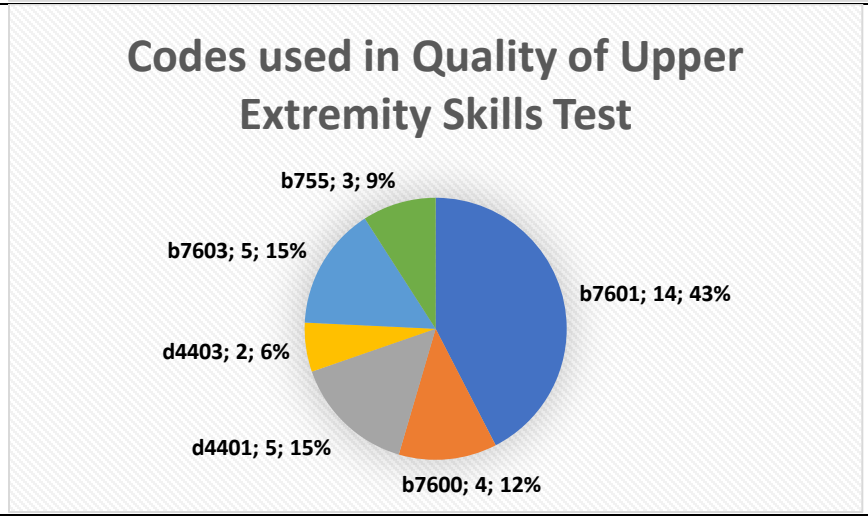
	releases behind the back to manipulate object	Capacity	N/A	N/A	release	manipulate	d4403 - Releasing	d4402 - Manipulating	
	Coordinating								
	coordinates grip using both hands	Capacity	N/A	N/A	coordinates	grip	b7602 - Coordination of voluntary movements	d4401 - Grasping	
	coordinates release using both hands	Capacity	N/A	N/A	coordinates	release	b7602 - Coordination of voluntary movements	d4401 - Grasping	
Upper Extremity Rating Scale (UERS)	Shoulder: active motion	Capacity	N/A	N/A	shoulder motion		b7100 - Mobility of a single joint		
	Elbow: active motion	Capacity	N/A	N/A	elbow motion		b7100 - Mobility of a single joint		
	Forearm: active motion (examine with elbow 45-90 degree)	Capacity	N/A	N/A	forearm motion		b7100 - Mobility of a single joint		
	Wrist: active motion (examine with elbow 45-90 degree)	Capacity	N/A	N/A	wrist motion		b7100 - Mobility of a single joint		
	Hand (grasp/release)	Capacity	N/A	N/A	grasp	release	d4401 - Grasping	d4403 - Releasing	
Test of Infant Motor Performance (TIMP)	*Observed items*								
	individual right finger movement	Capacity	N/A	N/A	finger movement		b7100 - Mobility of a single joint		
	individual left finger movement	Capacity	N/A	N/A	finger movement		b7100 - Mobility of a single joint		
	fingers object/surface on right	Capacity	N/A	N/A	fingers object/surface on right		d4452 - Reaching		
	fingers object/surface on left	Capacity	N/A	N/A	fingers object/surface on left		d4452 - Reaching		
	fidgety movement	Capacity	N/A	N/A	fidgety movement		b7611 - Specific spontaneous movements		

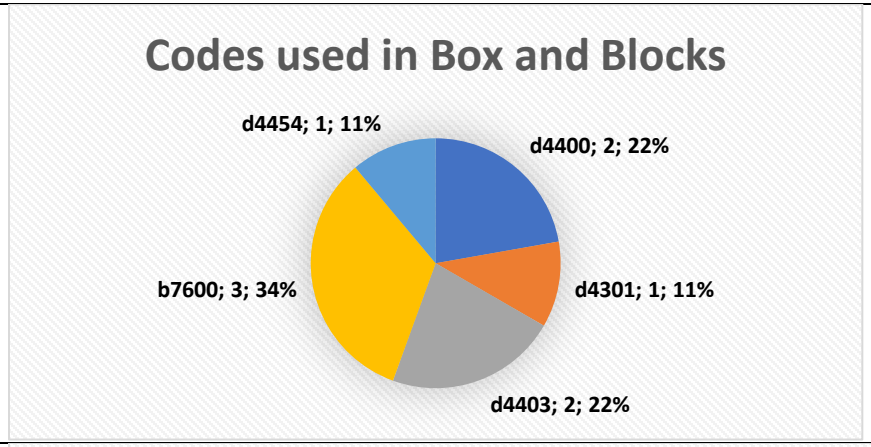
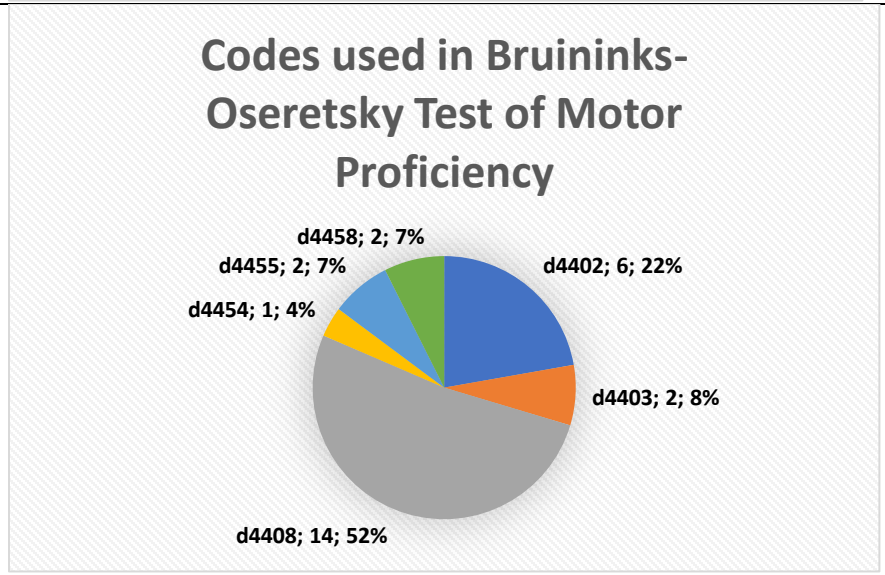
	ballistic movement of the arm or legs (swipes or swats)	Capacity	N/A	N/A	ballistic movement of the arm or legs (swipes or swats)		b7611 - Specific spontaneous movements		
	oscillation of arm or leg during movement	Capacity	N/A	N/A	oscillation of arm or leg during movement		b7611 - Specific spontaneous movements		
	Reaches for person or object	Capacity	N/A	N/A	reaches for person or object		d4452 - Reaching		
	Elicited items								
	Defensive reaction - arm movement	Capacity	N/A	N/A	defensive reaction - arm movement		b755 - Involuntary movement reaction functions		
	rolling: elicited from the arms	Capacity	N/A	N/A	rolling: elicited from the arms		d4107 - Rolling Over		
	pull to sit	Capacity	N/A	N/A	pull to sit		d4103 - Sitting		
	lateral straightening of the head and body with arm support	Capacity	N/A	N/A	supportive stability with arm		b7603 - Supportive functions of arm or leg		
	crawling	Capacity	N/A	N/A	crawling		d4450 - Pulling		
Video Observations Aarts and Aarts, module Determine Developmental Disregard (VOA-DDD)	Stringing beads (child was asked to take beads from six different cans and string them on a thread)	Capacity	N/A	N/A	stringing beads		d4402 - Manipulating		
	Decorating a muffin								
	Child was asked to put a placemat on the table and to put a place on the placemat	Capacity	N/A	N/A	placemat on table and put place on placemat		d6300 - Preparing simple meals		
	Child was asked to take the muffin from the saucer	Capacity	N/A	N/A	take the muffin from the saucer and put it on the		d6300 - Preparing simple meals		

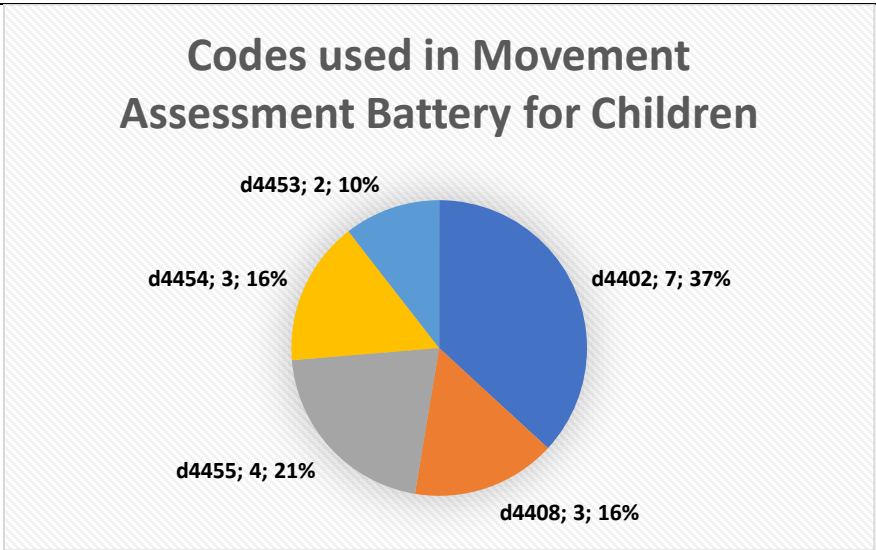
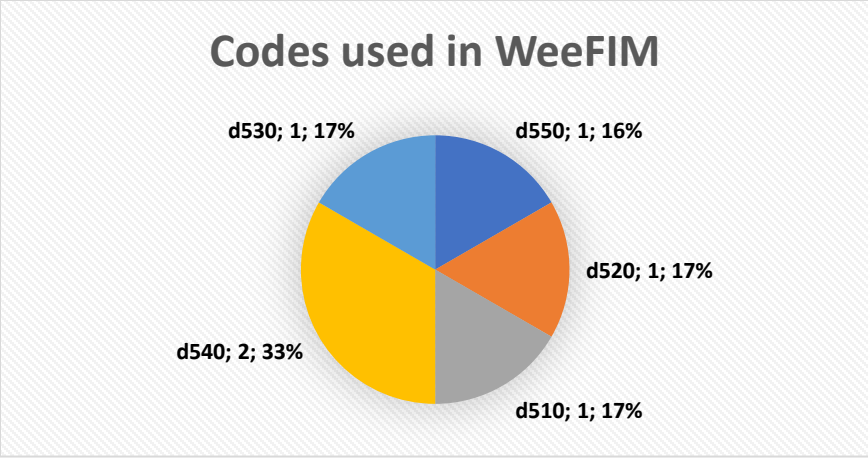
	and put it on the plate and to take the paper off the muffin and put the paper on the saucer				plate and to take the paper off the muffin and put the paper on the saucer				
	Child was asked to subsequently decorate the muffin by taking candies from the egg-cup and two pots and to put the caster sugar on the muffin	Capacity	N/A	N/A	decorate the muffin by taking candies from the egg-cup and two pots and to put the caster sugar on the muffin		d6300 - Preparing simple meals		
	For the older children, when decorating the muffin was finished, they were asked to cut the muffin into pieces	Capacity	N/A	N/A	they were asked to cut the muffin into pieces		d6300 - Preparing simple meals		
	Once completed they were asked to clean their hands with a napkin	Capacity	N/A	N/A	washing hands		d5100 - Washing body parts		

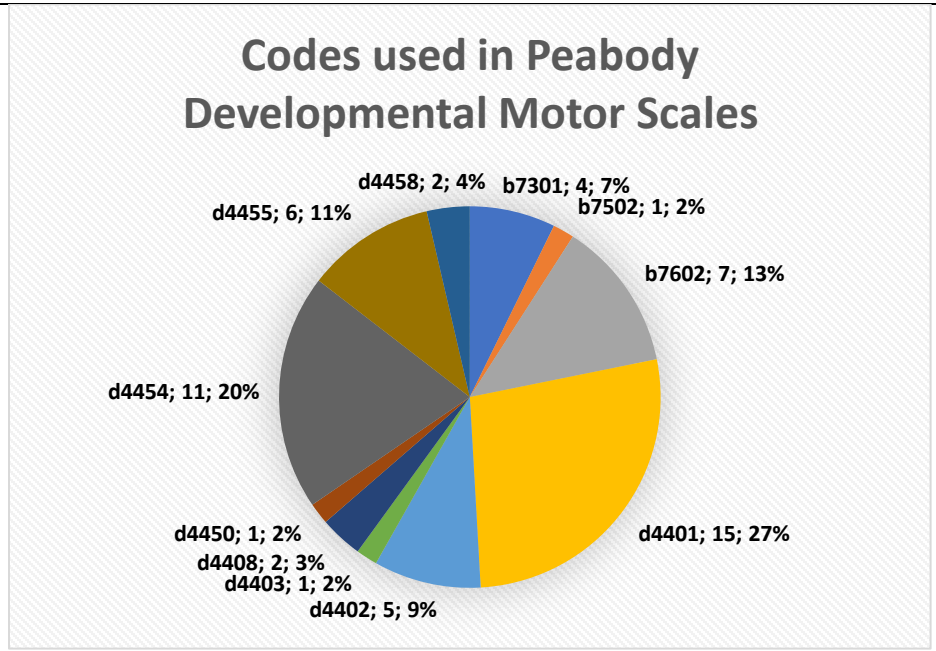
Appendix 2. Manuscript 1 - Table 5. Pie chart representation of codes used for each outcome measure.

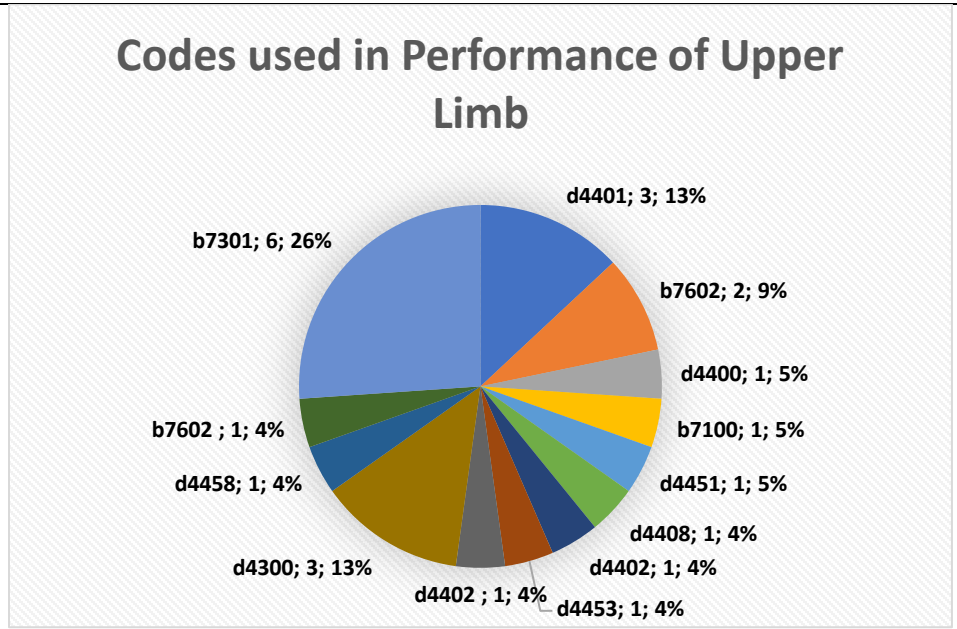
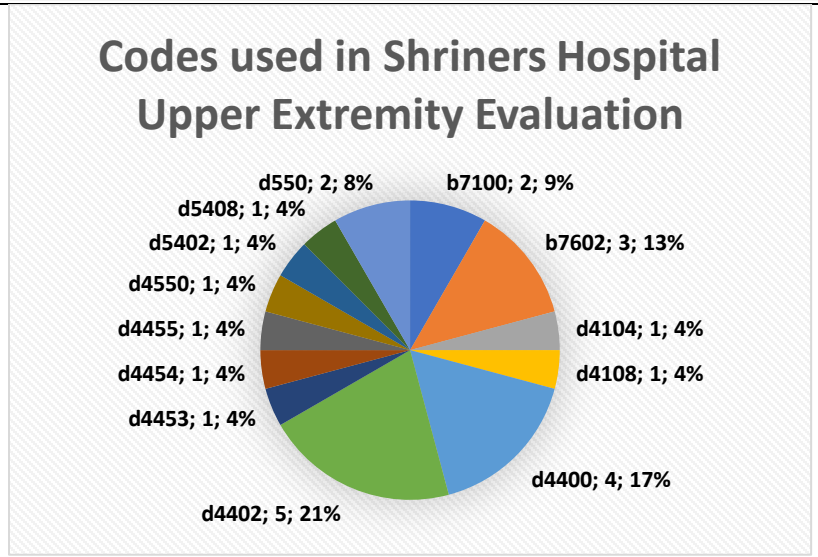
Outcome measure name	Pie Chart
Assisting Hand Assessment (AHA)	
Melbourne Assessment of Unilateral Upper Limb (MAUUL)	

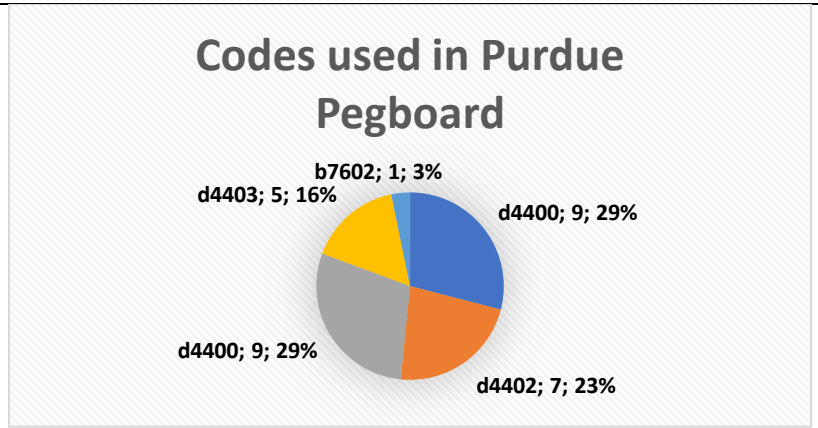
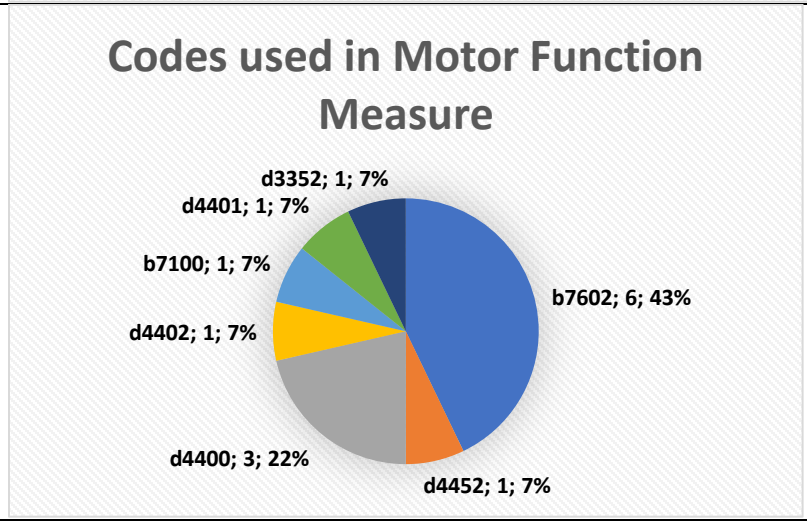
Jebsen-Taylor Hand Function Test	
Quality of Upper Extremity Skills Tests (QUEST)	

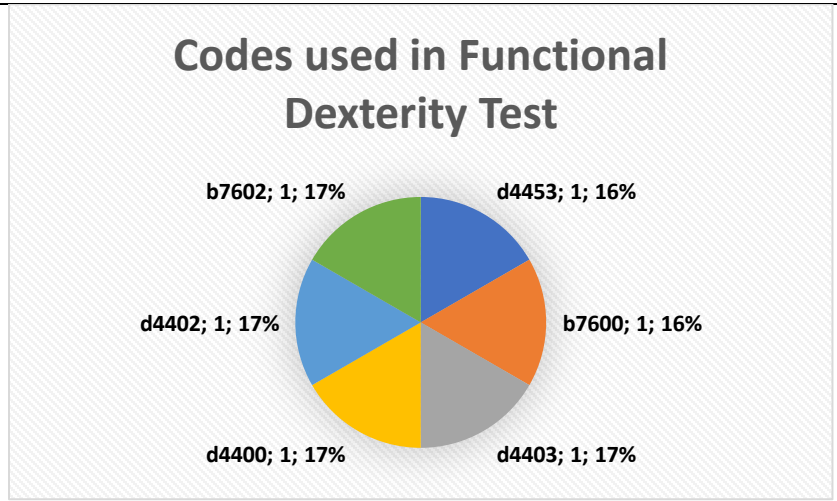
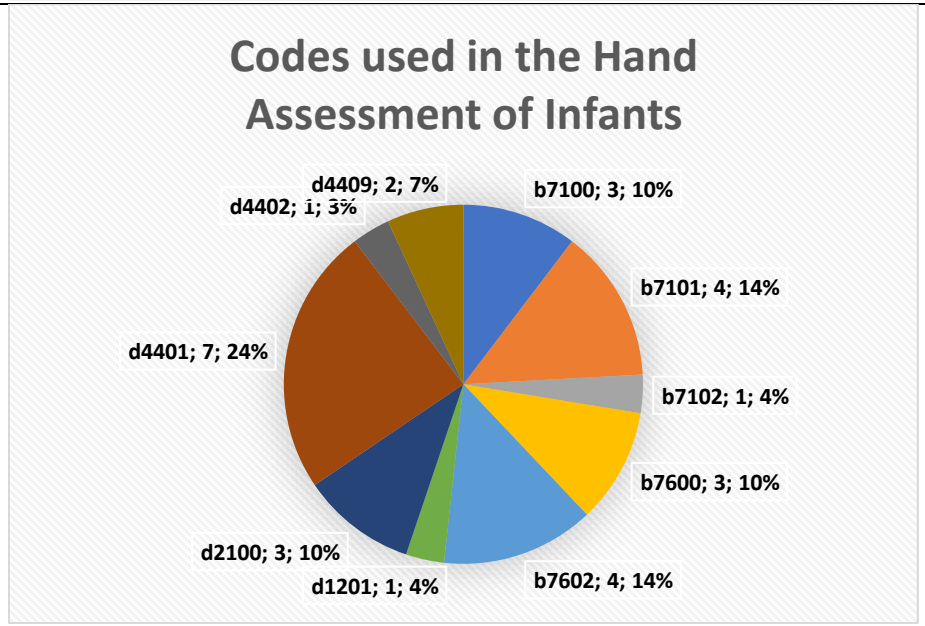
Box and Blocks	
Bruininks-Oseretsky (BOT-2)	

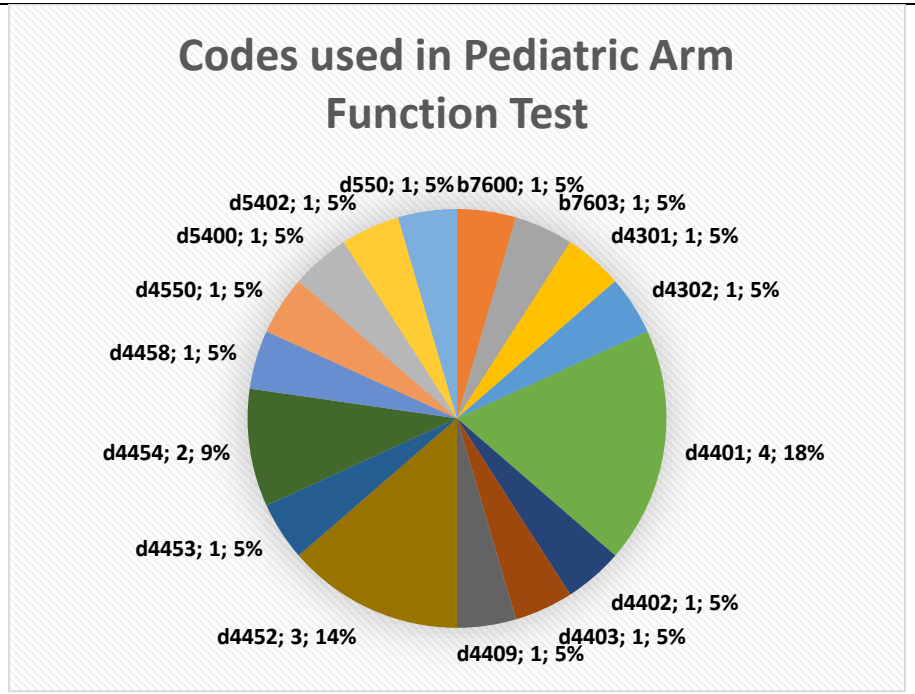
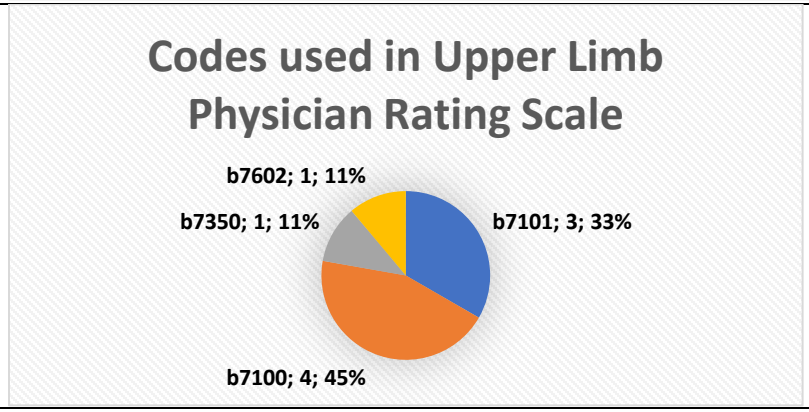
Movement Assessment Battery for Children (M-ABC)	
Functional Independence Measure for Children (WeeFIM)	

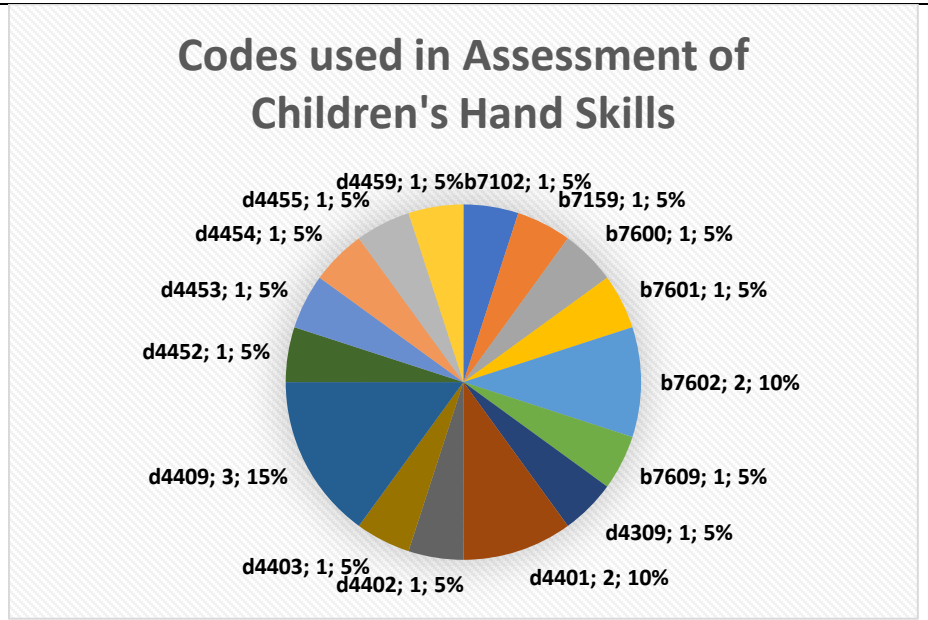
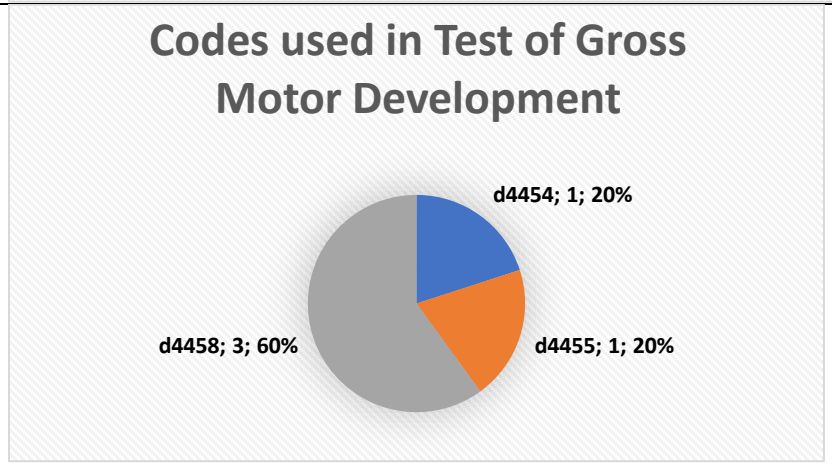
Peabody Developmental Motor Scales (PDMS)-2	

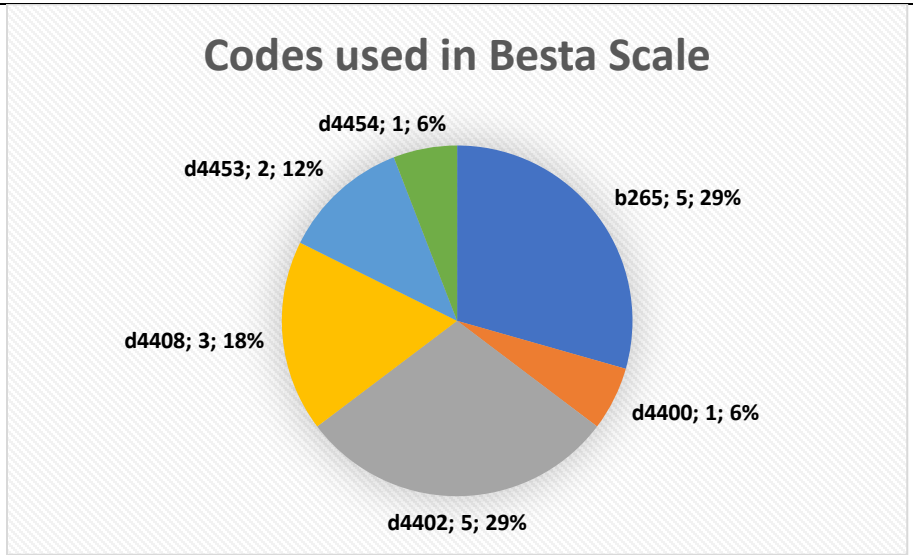
Performance of Upper Limb (PUL)	
Shriners Hospital for Children Upper Extremity Evaluation (SHUEE)	

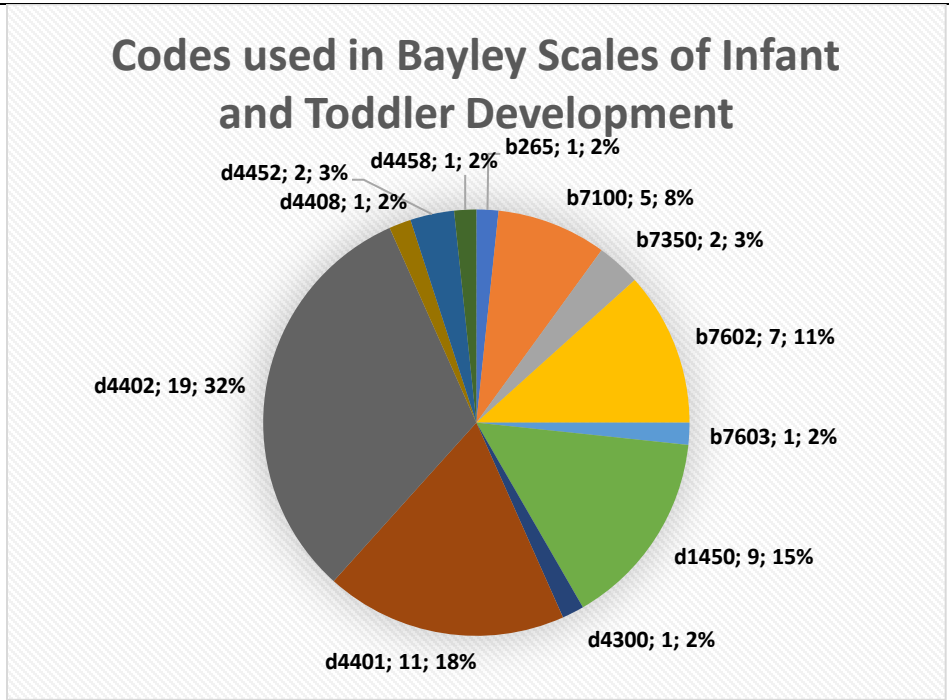
Purdue Pegboard Test	
Motor Function Measure (MFM)	

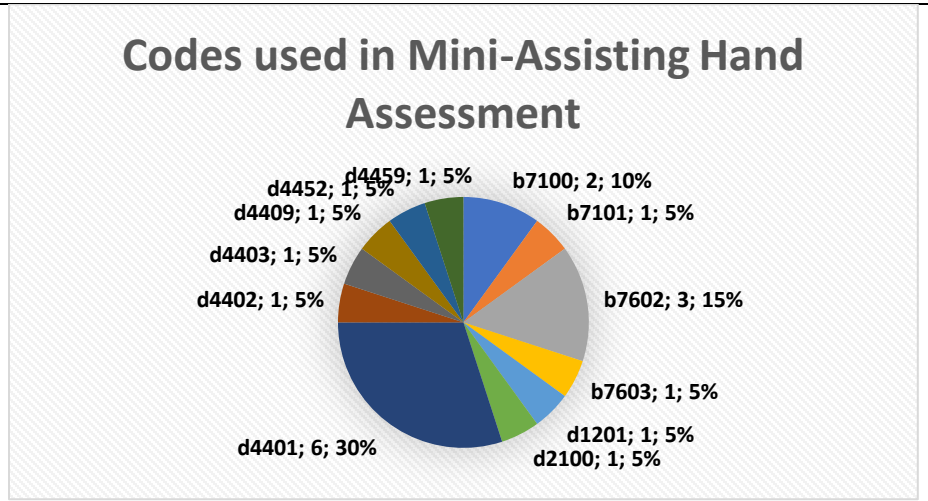
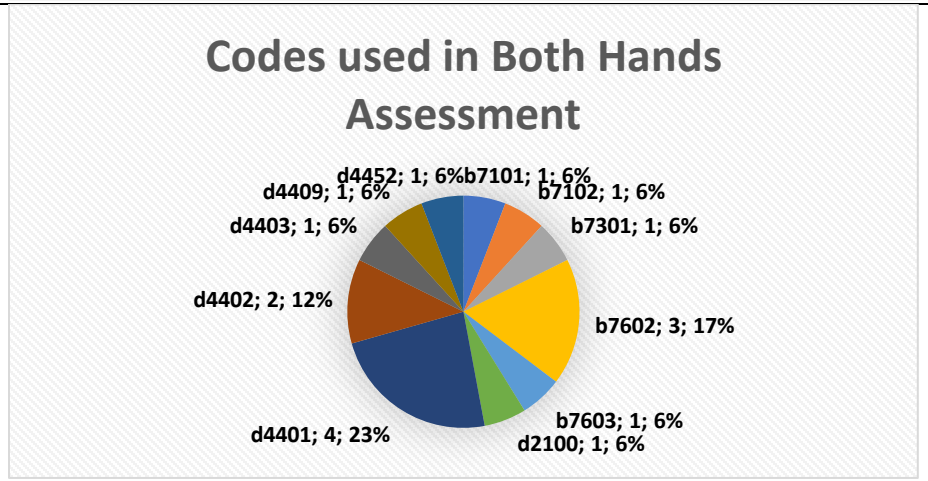
Functional Dexterity Test (FDT)	
Hand Assessment of Infants (HAI)	

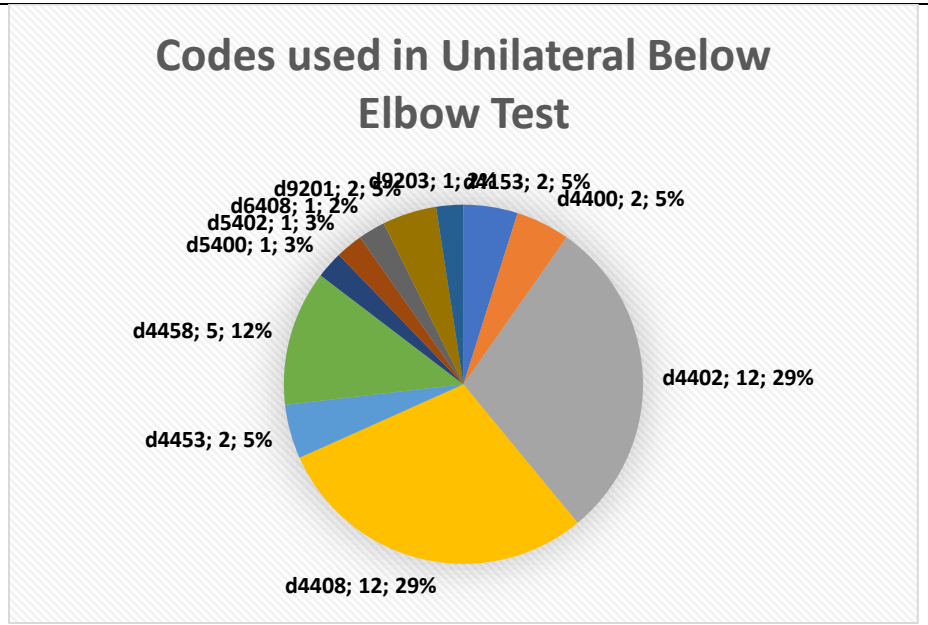
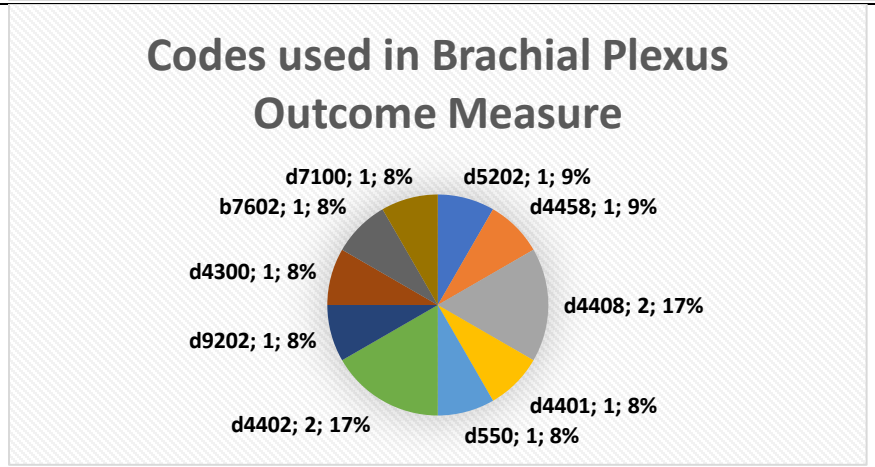
Pediatric Arm Function Test (PAFT)	
Upper Limb Physician Rating Scale (ULPRS)	

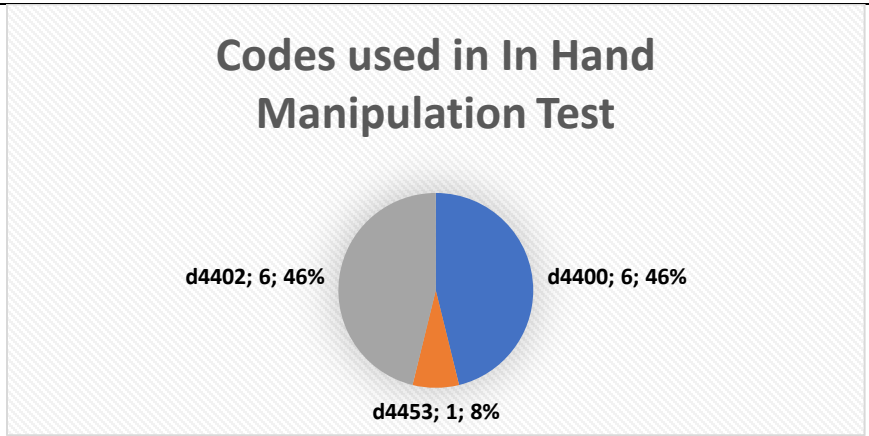
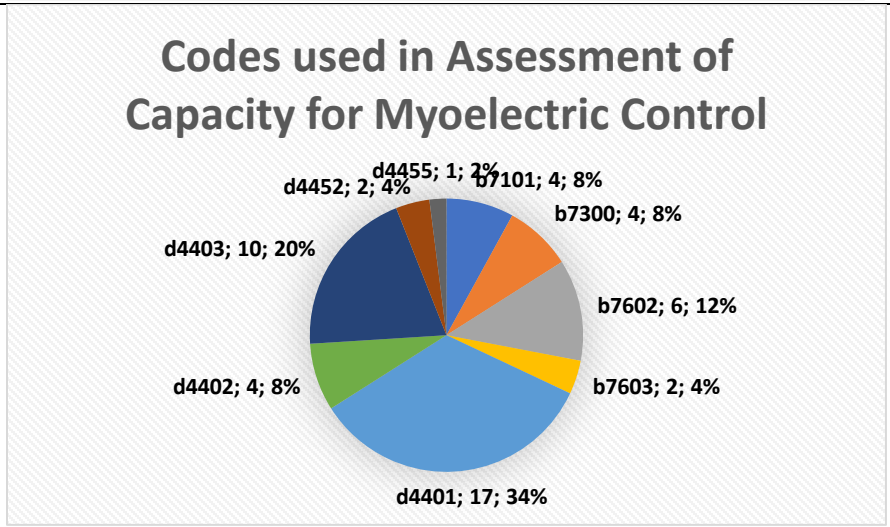
Assessment of Children Hand Skills (ACHS)	
Test of Gross Motor Development (TGMD) -2	

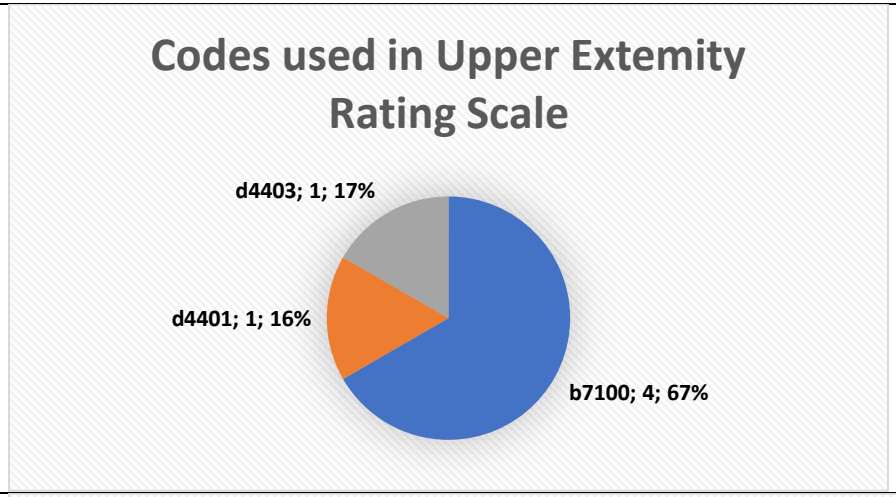
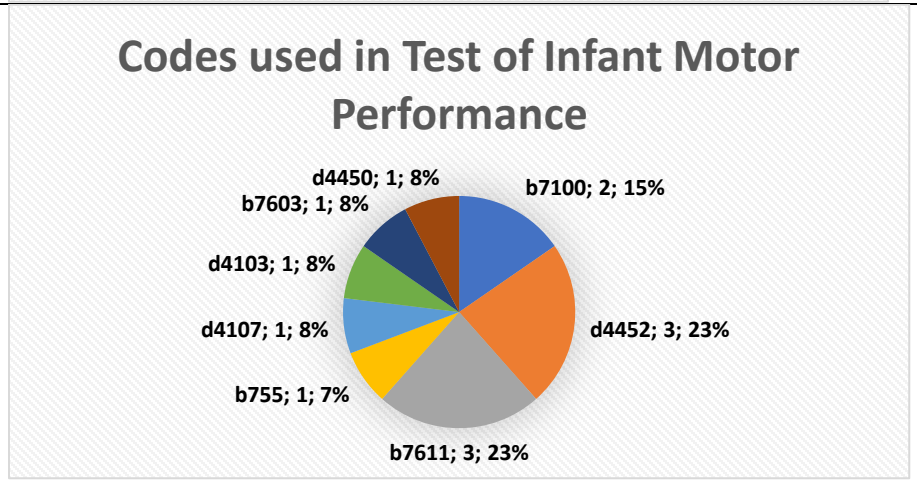
Besta Scale	
Selective Control of the Upper Extremity Scale (SCUES)	All 6 items were coded to b7601 - Control of complex voluntary movements

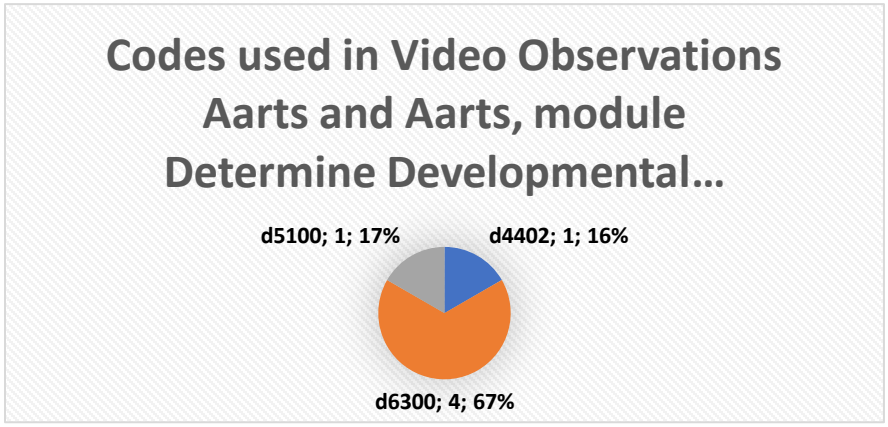
Bayley Scales of Infant and Toddler Development (BSID) - 3rd edition	
Minnesota Handwriting Assessment	only 1 item

Mini Assisting Hand Assessment (Mini AHA)	
Both Hands Assessment (BoHA)	

Unilateral Below Elbow Test (UBET)	
Brachial Plexus Outcome Measure (BPOM)	

In-Hand Manipulation Test	
Assessment of Capacity for Myoelectric Control (ACMC)	

Upper Extremity Rating Scale (UERS)	
Test of Infant Motor Performance (TIMP)	

Video Observations Aarts and Aarts, module Determine Developmental Disregard (VOA-DDD)	

Appendix 3. Manuscript 2 – Qualtrics Clinician Survey Round 1

Good day,

Our research team at the Shriners Hospital for Children - Canada is working on a project titled: **Development of the Shriners Hospitals Arthrogryposis Pediatric Evaluation - UPper Extremity (SHAPE-UP)**.
The purpose of the SHAPE-UP is to assess UE function in children with AMC with upper limb involvement. The SHAPE-UP will describe the impairments, activity limitations, and participation restrictions in the performance of daily tasks in children with AMC to guide treatment decision-making and evaluate treatment effectiveness for the UE.

We are inviting you to complete a survey that will help us gather information on test items specific to the UE. We will ask you some questions about your professional background and experience, as well as questions specific to test items when assessing children and youth with AMC. The survey takes approximately 20-30 minutes to complete.

By completing this survey, you consent to participate in the study. Please refer to the attached PDF for full consent discussion and details.

We thank you in advance for your participation and contribution to this project.

Please feel free to forward this questionnaire to other clinicians (occupational and physical therapists, physical rehabilitation technicians, physicians, and certified hand therapists) with 2 years or more of experience working with children and youth with AMC. Your help and support is very much appreciated.

If you have any questions regarding the survey, please contact the clinical research coordinator Kathleen Montpetit at kmontpetit@shrinenet.org

SHAPE-UP Purpose and Consent

Development of the Shriners Hospitals Arthrogryposis Pediatric Evaluation - UPper Extremity (SHAPE-UP)

You are being asked to take part in a research study because you are a clinician involved in the care of children with arthrogryposis multiplex congenita (AMC) with at least 2 years experience.

A major component in this process is to include the expertise of experienced practitioners worldwide, such as you. This survey will help us gather information about what are the most clinically important items to consider when evaluating the upper extremity (UE) function of children with AMC.

The purpose of the SHAPE-UP is to assess UE function in children with AMC with upper limb involvement. The SHAPE-UP will describe the impairments, activity limitations, and participation restrictions in the performance of daily tasks in children with AMC to guide treatment decision-making and evaluate treatment effectiveness for the UE.

Please read the consent form attached below. By proceeding with the survey, you acknowledge having read and understood the consent discussion and consent to participate in the study.

Click here: SHAPE-UP Consent Form

Demographic Information

What is your occupation?

- ○ Occupational Therapist
- ○ Physical Therapist
- ○ Physician
- ○ Certified Hand Therapist
- ○ Physical Rehabilitation Technician (TRP)
- ○ _____ Other (Please specify)

What specialty of medicine do you practice?

In what country do you currently work?

In which type of setting are you currently working?

- ☐ Hospital
- ☐ Community Center
- ☐ Rehabilitation Center
- ☐ Private Center/Private Practice
- ☐ School-Based Practice
- ☐ _____ Other (Please Specify)

Do you have more than 2 years experience working with children/youth (1-21 years) with AMC?

- ○ Yes
- ○ No

How many years have you been working with children/youth (1-21 years) with AMC?

How many children/youth with AMC have you worked with?

Which healthcare professionals do you work with when caring for children with AMC? Choose all that apply:

- ☐ Pediatrician
- ☐ Orthopedic Surgeon
- ☐ Nurse
- ☐ Orthotist
- ☐ Occupational Therapist
- ☐ Physical Therapist
- ☐ Physiatrist
- ☐ Geneticist
- ☐ Neurologist
- ☐ Social Worker
- ☐ Psychologist
- ☐ Speech Language Pathologist
- ☐ _____ Other (Please Specify)

Elements of the SHAPE-UP: ROM

Which movement of the **SHOULDER** do you feel is necessary to screen for UE function? (Choose all that apply)

- ☐ ADduction
- ☐ ABduction
- ☐ Flexion
- ☐ Extension
- ☐ Internal rotation
- ☐ External rotation
- ☐ _____ Other (Please specify)

Which movement of the **ELBOW** do you feel is necessary to screen for UE function? (Choose all that apply)

- ☐ Extension
- ☐ Flexion

Which movement of the **FOREARM** do you feel is necessary to screen for UE function? (Choose all that apply)

- ☐ Pronation
- ☐ Supination

Which movement of the **WRIST** do you feel is necessary to screen for UE function? (Choose all that apply)

- ☐ Flexion
- ☐ Extension
- ☐ Ulnar deviation
- ☐ Radial deviation

Which movement of the **FINGERS** do you feel is necessary to screen for UE function? (Choose all that apply)

- ☐ Flexion
- ☐ Extension
- ☐ ABduction

☐ ADduction
☐ Other (Please specify)

Which movement of the **THUMB** do you feel is necessary to screen for UE function? (Choose all that apply)

☐ Flexion
☐ Extension
☐ ADduction
☐ ABduction
☐ Opposition

Elements of the SHAPE-UP: Scoring

How should the items of the SHAPE-UP be scored? Please rank your response by dragging and dropping in the order your prefer.

Timed

Level of assistance
0 – Unable to complete
1 – Able to complete with human assistance
2 – Able to complete with adaptive equipment
3 – Able to complete independently

Level of performance (ability to complete the task)

Qualifying the position of the limb segments for the specific item (e.g. positional analysis in the SHUEE)

Other (Please specify)

Please provide any additional comments regarding scoring options.

Elements of the SHAPE-UP: Items

For the Item Block of this questionnaire, please keep in mind that the following items are **performance-based**. This means that we want the child with AMC to perform these items in the setting while the clinician observes them complete the task.

The items fall under 11 subdomains. These domains are:
- Upper Extremity Weight Bearing
- Using the Upper Extremity to change Body Position
- Grasp
- Release
- Reach
- Fine Hand Manipulation
- Throwing
- Catching
- Dressing
- Feeding
- Toileting

For the domain **"Upper Extremity Weight Bearing"** please rate the following items for inclusion in the SHAPE-UP measure as well as add any additional comments. These could include changing the wording, adding a missing item, or changing the meaning of the item.

	Essential	Useful but not essential	Not necessary	Comment
Uses non-dominant hand to stabilize self	☐	☐	☐	☐
Uses upper extremity to weight bear in prone	☐	☐	☐	☐
Uses upper extremity to weight bear in sitting with hands forward	☐	☐	☐	☐
Uses upper extremity to weight bear in sitting with hands to the side	☐	☐	☐	☐
Uses upper extremity to weight bear in sitting with hands behind body	☐	☐	☐	☐

Please provide any additional comments for the domain **"Upper Extremity Weight Bearing"**

For the domain **"Using the Upper Extremity to Change Body Position"** please rate the following items for inclusion in the SHAPE-UP measure as well as add any additional comments. These could include changing the wording, adding a missing item, or changing the meaning of the item.

	Essential	Useful but not essential	Not necessary	Comment
Get down and crawl towards me	☐	☐	☐	☐
Pushes self from supine to sitting	☐	☐	☐	☐
Pushes self from sitting to standing	☐	☐	☐	☐
Transferring from one body position to another	☐	☐	☐	☐
Pushes self using upper extremity (bum scoot)	☐	☐	☐	☐
Using upper extremity to use mobility aid	☐	☐	☐	☐

Please provide any additional comments for the domain **"Using the Upper Extremity to Change Body Position"**

For the domain **"Grasp"** please rate the following items for inclusion in the SHAPE-UP measure as well as add any additional comments. These could include changing the wording, adding a missing item, or changing the meaning of the item.

	Essential	Useful but not essential	Not necessary	Comment
Picking up a large object (box, backpack, jacket, etc.)	☐	☐	☐	☐
Picking up a medium object (shoes, water bottle, plate, etc.)	☐	☐	☐	☐
Picking up a small object (coin, bead, pencil, etc.)	☐	☐	☐	☐

Please provide any additional comments for the domain **"Grasp"**

For the domain **"Release"** please rate the following items for inclusion in the SHAPE-UP measure as well as add any additional comments. These could include changing the wording, adding a missing item, or changing the meaning of the item.

	Essential	Useful but not essential	Not necessary	Comment
Releasing a large object (box, backpack, jacket, etc.)	☐	☐	☐	☐
Releasing a medium object (shoes, water bottle, plate, etc.)	☐	☐	☐	☐
Releasing a small object (coin, bead, pencil, etc.)	☐	☐	☐	☐

Please provide any additional comments for the domain **"Release"**

For the domain **"Reach"** please rate the following items for inclusion in the SHAPE-UP measure as well as add any additional comments. These could include changing the wording, adding a missing item, or changing the meaning of the item.

	Essential	Useful but not essential	Not necessary	Comment
Reaching for an item from floor level	☐	☐	☐	☐
Reaching for an item from waist level	☐	☐	☐	☐
Reaching for an item overhead	☐	☐	☐	☐
Reaching for an item by crossing the midline	☐	☐	☐	☐

Please provide any additional comments for the domain **"Reach"**

For the domain **"Fine Hand Manipulation"** please rate the following items for inclusion in the SHAPE-UP measure as well as add any additional comments. These could include changing the wording, adding a missing item, or changing the meaning of the item.

	Essential	Useful but not essential	Not necessary	Comment
Tearing a sheet of paper into many pieces	☐	☐	☐	☐
Cutting geometric shapes (circle, square, triangle)	☐	☐	☐	☐
Using scissors to snip the paper	☐	☐	☐	☐
Stringing beads together	☐	☐	☐	☐
Folding a piece of paper	☐	☐	☐	☐
Stacking blocks	☐	☐	☐	☐
Opening a band-aid	☐	☐	☐	☐
Write your name or draw something	☐	☐	☐	☐
Opening a jar	☐	☐	☐	☐
Closing a jar	☐	☐	☐	☐
Turning over a card	☐	☐	☐	☐

Please provide any additional comments for the domain **"Fine Hand Manipulation"**

For the domain **"Throwing"** please rate the following items for inclusion in the SHAPE-UP measure as well as add any additional comments. These could include changing the wording, adding a missing item, or changing the meaning of the item.

	Essential	Useful but not essential	Not necessary	Comment
Throwing a ball overhead	☐	☐	☐	☐
Throwing a ball underhand	☐	☐	☐	☐

Please provide any additional comments for the domain **"Throwing"**

For the domain **"Catching"** please rate the following items for inclusion in the SHAPE-UP measure as well as add any additional comments. These could include changing the wording, adding a missing item, or changing the meaning of the item.

	Essential	Useful but not essential	Not necessary	Comment
Catching a ball overhead one handed	☐	☐	☐	☐
Catching a ball underhand one handed	☐	☐	☐	☐
Catching a ball overhead two handed	☐	☐	☐	☐
Catching a ball underhand two handed	☐	☐	☐	☐

Please provide any additional comments for the domain **"Catching"**

For the domain **"Dressing"** please rate the following items for inclusion in the SHAPE-UP measure as well as add any additional comments. These could include changing the wording, adding a missing item, or changing the meaning of the item.

	Essential	Useful but not essential	Not necessary	Comment
Puts on clothes over-head (shirt, sweater, hat)	☐	☐	☐	☐
Puts on open shirt	☐	☐	☐	☐
Removes shirt	☐	☐	☐	☐
Buttoning	☐	☐	☐	☐

	Essential	Useful but not essential	Not necessary	Comment
Unbuttoning	☐	☐	☐	☐
Starts a zipper	☐	☐	☐	☐
Pull up zipper	☐	☐	☐	☐
Pulls down zipper	☐	☐	☐	☐
Snaps fasteners	☐	☐	☐	☐
Unsnaps fasteners	☐	☐	☐	☐
Pulls on pants	☐	☐	☐	☐
Pulls down (remove) pants	☐	☐	☐	☐
Puts on socks	☐	☐	☐	☐
Remove socks	☐	☐	☐	☐
Puts on shoes	☐	☐	☐	☐
Ties shoelaces	☐	☐	☐	☐
Removes shoes	☐	☐	☐	☐

Please provide any additional comments for the domain **"Dressing"**

For the domain **"Feeding"** please rate the following items for inclusion in the SHAPE-UP measure as well as add any additional comments. These could include changing the wording, adding a missing item, or changing the meaning of the item.

	Essential	Useful but not essential	Not necessary	Comment
Reaches mouth	☐	☐	☐	☐
Cuts food using knife	☐	☐	☐	☐
Picks up food using fork	☐	☐	☐	☐

Please provide any additional comments for the domain **"Feeding"**

For the domain **"Toileting"** please rate the following items for inclusion in the SHAPE-UP measure as well as add any additional comments. These could include changing the wording, adding a missing item, or changing the meaning of the item.

	Essential	Useful but not essential	Not necessary	Comment
Places sticker on lower back (proxy for reaching to wipe buttocks)	☐	☐	☐	☐

Please provide any additional comments for the domain **"Toileting"**

Additional Comments

Please add any comments you may have regarding any additional items you feel should be included in the SHAPE-UP measure that were not included in the survey.

Appendix 4. Manuscript 2 – Qualtrics Clinician Survey Round 2

The purpose of the SHAPE-UP is to assess upper extremity (UE) function in children with AMC with upper limb involvement. The SHAPE-UP will describe the impairments, activity limitations, and participation restrictions in the performance of daily tasks in children with AMC to guide treatment decision-making and evaluation treatment effectiveness for the UE.

In the tasks provided below, this will be the scoring guide implemented for the SHAPE-UP:

0 = Unable - The child is unable to complete any component of the task.
1 = **Partial** completion of task **passively** - The child can partially complete the task using passive range of motion.
2 = **Partial** completion of task **actively** - The child can partially compelte the task using active range of motion.
3 = **Completion** of task **passively** - The child can complete the task using passively range of motion.
4 = **Completion** of task **actively** - The child can complete the task using active range of motion.

Block 0 - Introduction

Thank you for participating in the SHAPE-UP Delphi Round #2. You are being invited to participate as you have previously completed round 1 of the survey. We have since taken into account all comments and feedback and have created a first version of the SHAPE-UP measure that we will present shortly.

Please note there are 12 tasks in the SHAPE-UP measure as well as 3 descriptive questions. The purpose of the measure as well as the scoring grid will be provided throughout the survey.

Please download the following PDF as a reference for the scoring tables if you require them.
LINK

We appreciate your time and efforts in completing this survey.

Block 1

Do you have any comments regarding the following task:

Task #1: Grasp/pick up a cheerio, bring it to your mouth, place it back down in front of you, and release it/let it go.

Grasp/pick up a cheerio	
bring it to your mouth	
place it back down in front of you	
release it/let it go	

Which word do you prefer?

- ○ Grasp a cheerio
- ○ Pick up a cheerio

Which word do you prefer?

- ○ release it
- ○ let it go

For the following task, we would like you to comment if an additional joint should be analyzed. We believe this task is focused more on the ***Thumb, Fingers, Elbow***.

Task #1: ${q://QID9/ChoiceGroup/SelectedChoices} a cheerio, bring it to your mouth, place it back down in front of you, and ${q://QID17/ChoiceGroup/SelectedChoices}.

Item	Score					Asst. Device	Arm		Analysis of Joint Motion and Position					
							R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Grasp/ Pick up	0	1	2	3	4				Palm/close/open	Flexion/neutral/extension	Flexion/neutral/extension	-	-	-
Bring to mouth	0	1	2	3	4				-	-	-	Pronated/ /neutral/supination	Extension/neutral/flexion	-
Bring back to table	0	1	2	3	4				-	-	-	Supination/neutral/ pronation	Flexion/neutral/extension	-
Release/let go	0	1	2	3	4				Palm/close/open	Flexion/neutral/extension	Flexion/neutral/extension	-	-	-

Remember that for the Analysis of Joint Motion and Position, the scoring options represent less to more function/optimal position for the item in question.

Please select which joint(s) should be **targeted** in the scoring sheet for the specified task.

	Thumb Score		Fingers Score		Wrist Score		Forearm Score		Elbow Score		Shoulder Score	
	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No

	Thumb Score		Fingers Score		Wrist Score		Forearm Score		Elbow Score		Shoulder Score	
	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No
Grasp/ Pick up	○	○	○	○	○	○	○	○	○	○	○	○
Bring to mouth	○	○	○	○	○	○	○	○	○	○	○	○
Bring back to table	○	○	○	○	○	○	○	○	○	○	○	○
Release/ let go	○	○	○	○	○	○	○	○	○	○	○	○

Other thoughts or notes regarding Task #1: ${q://QID9/ChoiceGroup/SelectedChoices} a cheerio, bring it to your mouth, place it back down in front of you, and ${q://QID17/ChoiceGroup/SelectedChoices}.

Block 2

Do you have any comments regarding the following task:

Task #2: Grasp/pick up a water bottle/can, bring it to your mouth, place it back down in front of you, and release it/let it go.

Grasp/pick up a water bottle/can

bring it to your mouth

place it back down in front of you

release it/let it go

Which word do you prefer?

○ Grasp

○ Pick up

Which word do you prefer?

- ○ water bottle
- ○ can

Which word do you prefer?

- ○ release it
- ○ let it go

For the following task, we would like you to comment if an additional joint should be analyzed. We believe this task is focused more on the ***Thumb, Fingers, Elbow***.

Task #2: ${q://QID13/ChoiceGroup/SelectedChoices}
a ${q://QID18/ChoiceGroup/SelectedChoices}, bring it to your mouth, place it back down in front of you, and ${q://QID19/ChoiceGroup/SelectedChoices}.

Item	Score					Asst. Device	Arm		Analysis of Joint Motion and Position					
							R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Grasp/ Pick up	0	1	2	3	4				Palm/close/open	Flexion/neutral/extension	Flexion/neutral/extension	-	-	-
Bring to mouth	0	1	2	3	4				-	-	-	Pronated/ /neutral/supination	Extension/neutral/flexion	-
Bring back to table	0	1	2	3	4				-	-	-	Supination/neutral/ pronation	Flexion/neutral/extension	-
Release/let go	0	1	2	3	4				Palm/close/open	Flexion/neutral/extension	Flexion/neutral/extension	-	-	-

Remember that for the Analysis of Joint Motion and Position, the scoring options represent less to more function/optimal position for the item in question.

Please select which joint(s) should be **targeted** in the scoring sheet for the specified task.

	Thumb Score		Fingers Score		Wrist Score		Forearm Score		Elbow Score		Shoulder Score	
	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No
Grasp/ Pick up	○	○	○	○	○	○	○	○	○	○	○	○
Bring to mouth	○	○	○	○	○	○	○	○	○	○	○	○
Bring back to table	○	○	○	○	○	○	○	○	○	○	○	○
Release/ let go	○	○	○	○	○	○	○	○	○	○	○	○

Other thoughts or notes regarding Task #2: ${q://QID13/ChoiceGroup/SelectedChoices}
a ${q://QID18/ChoiceGroup/SelectedChoices}, bring it to your mouth, place it back down in front of you, and ${q://QID19/ChoiceGroup/SelectedChoices}.

Block 3

Do you have any comments regarding the following task:

Task #3: Open the jar, pour out a few beads/macaroni/buttons, string 2 or 3 together, and close the jar [small size jar e.g. spice jar or salt/paper shaker]

Open the jar

Pour out a few beads/macaroni/buttons

String 2 or 3 together

Close the jar

Which word do you prefer?

- ○ beads
- ○ macaroni
- ○ buttons

For the following task, we would like you to comment if an additional joint should be analyzed. We believe this task is focused more on the ***Thumb, Fingers, Wrist***.

Task #3: Open the jar, pour out a few ${q://QID21/ChoiceGroup/SelectedChoices}, string 2 or 3 together, and close the jar.

Item	Score					Asst. Device	Arm		Analysis of Joint Motion and Position					
							R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Stabilizes jar	0	1	2	3	4				Palm/close/open	No finger movement/partial finger movement/ full finger movement	Extension/ flexion/neutral	Supination/pronation/ neutral	-	-
Open jar	0	1	2	3	4				Palm/close/open	No finger movement/partial finger movement/ full finger movement	Extension/ Neutral/ flexion	Supination/neutral/ pronation	-	-
Pour out	0	1	2	3	4				Palm/close/open	-	-	Supination/neutral/ pronation	-	-
Hold small item	0	1	2	3	4				Palm/close/open	Fist/Finger adduction/True opposition	-	-	-	-
String	0	1	2	3	4				Palm/close/open	Fist/Finger adduction/True opposition	-	-	-	-
Close jar	0	1	2	3	4				Palm/close/open	No finger movement/partial finger movement/ full finger movement	Extension/ Neutral/ flexion	Supination/neutral/ pronation	-	-

Remember that for the Analysis of Joint Motion and Position, the scoring options represent less to more function/optimal position for the item in question.

Please select which joint(s) should be **targeted** in the scoring sheet for the specified task.

	Thumb Score		Fingers Score		Wrist Score		Forearm Score		Elbow Score		Shoulder Score	
	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No
Stabilizes jar	○	○	○	○	○	○	○	○	○	○	○	○
Open jar	○	○	○	○	○	○	○	○	○	○	○	○
Pour out	○	○	○	○	○	○	○	○	○	○	○	○
Hold small item	○	○	○	○	○	○	○	○	○	○	○	○
String	○	○	○	○	○	○	○	○	○	○	○	○
Close jar	○	○	○	○	○	○	○	○	○	○	○	○

Other thoughts or notes regarding Task #3: Open the jar, pour out a few ${q://QID21/ChoiceGroup/SelectedChoices}, string 2 or 3 together, and close the jar.

Block 4

Do you have any comments regarding the following task:

Task #4: Pick up the crayon/marker, write your name on this piece of paper, fold the paper, and cut it using the scissors. [Therapist to place crayon/marker and scissors across the midline]

Pick up the crayon/marker	
Write your name on this piece of paper	
Fold the paper	
Cut it using the scissors	

Which word do you prefer?

- ○ crayon
- ○ marker

For the following task, we would like you to comment if an additional joint should be analyzed. We believe this task is focused more on the ***Thumb, Fingers, Wrist***.

Task #4: Pick up the ${q://QID28/ChoiceGroup/SelectedChoices}, write your name on this piece of paper, fold the paper, and cut it using the scissors

Item	Score					Asst. Device	Arm		Analysis of Joint Motion and Position					
							R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
pick up	0	1	2	3	4				Palm/close/open	No finger movement/partial finger movement/ full finger movement	Flexion/neutral/ extension	supination/neutral/pronation	-	-
write name	0	1	2	3	4				Palm/close/open	No finger movement/partial finger movement/ full finger movement	Flexion/neutral/ extension	supination/neutral/pronation	-	-
fold paper	0	1	2	3	4				Palm/close/open	No finger movement/partial finger movement/ full finger movement	Flexion/neutral/ extension	-	-	-
cut	0	1	2	3	4				Palm/close/open	No finger movement/partial finger movement/ full finger movement	Flexion/neutral/ extension	supination/pronation/neutral	-	-
stabilize paper to cut	0	1	2	3	4				Palm/close/open	No finger movement/partial finger movement/ full finger movement	Flexion/neutral/ extension	supination/pronation/neutral	-	-

Remember that for the Analysis of Joint Motion and Position, the scoring options represent less to more function/optimal position for the item in question.

Please select which joint(s) should be **targeted** in the scoring sheet for the specified task.

	Thumb Score		Fingers Score		Wrist Score		Forearm Score		Elbow Score		Shoulder Score	
	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No
Pick up	○	○	○	○	○	○	○	○	○	○	○	○
Write name	○	○	○	○	○	○	○	○	○	○	○	○
Fold paper	○	○	○	○	○	○	○	○	○	○	○	○
Cut	○	○	○	○	○	○	○	○	○	○	○	○
Stabilize paper to cut	○	○	○	○	○	○	○	○	○	○	○	○

Other thoughts or notes regarding Task #4: Pick up the ${q://QID28/ChoiceGroup/SelectedChoices}, write your name on this piece of paper, fold the paper, and cut it using the scissors.

Block 5

Do you have any comments regarding the following task:

Task #5: Pick up the Play-Doh using the fork and bring it to your mouth. [Therapist to place items in front of child]

Pick up the Play-Doh using the fork

Bring it to your mouth

For the following task, we would like you to comment if an additional joint should be analyzed. We believe this task is focused more on the ***Wrist and Forearm***.

Task #5: Pick up the Play-Doh using the fork and bring it to your mouth.

Item	Score	Asst. Device	Arm		Analysis of Joint Motion and Position					
			R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Pick up the play doh	0 1 2 3 4				palm/close/open	Fist /Finger adduction/True opposition	flexion/extension/neutral	supination/pronation/neutral	-	-
bring fork to mouth	0 1 2 3 4				-	-	-	Pronation//neutral/supination	Extension/neutral/flexion	-

Remember that for the Analysis of Joint Motion and Position, the scoring options represent less to more function/optimal position for the item in question.

Please select which joint(s) should be **targeted** in the scoring sheet for the specified task.

	Thumb Score		Fingers Score		Wrist Score		Forearm Score		Elbow Score		Shoulder Score	
	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No
Pick up the Play-Doh	○	○	○	○	○	○	○	○	○	○	○	○
Bring fork to mouth	○	○	○	○	○	○	○	○	○	○	○	○

Other thoughts or notes regarding Task #5: Pick up the Play-Doh using the fork and bring it to your mouth.

Block 6

Do you have any comments regarding the following task:

Task #6: Reach for a small-size (e.g., tennis ball) ball placed on the floor, throw the ball underhand. Repeat task, throwing ball overhead.

Reach for small-size ball placed on the floor	
throw the ball underhand	
Repeat task	
Throwing ball overhead	

For the following task, we would like you to comment if an additional joint should be analyzed. We believe this task is focused more on the ***Elbow and Shoulder***.

Task #6: Reach for a small-size (e.g., tennis ball) ball placed on the floor, throw the ball underhand. Repeat task, throwing ball overhead.

Item	Score					Asst. Device	Arm		Analysis of Joint Motion and Position					
							R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Reach ball on floor	0	1	2	3	4				-	-	Flexion/neutral/ extension	Supination/ neutral/pronation	Flexion/neutral /extension	Extension/neutral/ flexion
throw ball underhand (winding/extend back at shoulder)	0	1	2	3	4				-	-	Flexion/neutral/ extension	Supination/ neutral/pronation	Flexion/neutral /extension	Flexion/neutral/ extension
throw ball underhand (release at shoulder)	0	1	2	3	4						Flexion/neutral/ extension	Supination/ neutral/pronation	Flexion/neutral /extension	Extension/neutral/ flexion
throw ball overhand	0	1	2	3	4				-	-	Flexion/neutral/ extension	Pronation/neutral/ supination	Flexion/neutral /extension	Extension/neutral/ flexion

Remember that for the Analysis of Joint Motion and Position, the scoring options represent less to more function/optimal position for the item in question.

Please select which joint(s) should be **targeted** in the scoring sheet for the specified task.

	Thumb Score		Fingers Score		Wrist Score		Forearm Score		Elbow Score		Shoulder Score	
	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No
Reach ball on floor	○	○	○	○	○	○	○	○	○	○	○	○

	Thumb Score		Fingers Score		Wrist Score		Forearm Score		Elbow Score		Shoulder Score	
	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No
Throw ball underhand (winding/extend at shoulder)	○	○	○	○	○	○	○	○	○	○	○	○
Throw ball underhand (release at shoulder)	○	○	○	○	○	○	○	○	○	○	○	○
Throw ball overhead	○	○	○	○	○	○	○	○	○	○	○	○

Other thoughts or notes regarding Task #6: Reach for a small-size (e.g., tennis ball) ball placed on the floor, throw the ball underhand. Repeat task, throwing ball overhead.

Block 7

Do you have any comments regarding the following task:

Task #7: Reach for a medium-sized (e.g., basket ball) ball placed on the floor, throw the ball underhand. Repeat task, throwing ball overhead.

Reach for medium-sized ball placed on the floor

throw the ball underhand

Repeat task

Throwing ball overhead

For the following task, we would like you to comment if an additional joint should be analyzed. We believe this task is focused more on the ***Elbow and Shoulder***.

Task #7: Reach for a medium-sized (e.g., tennis ball) ball placed on the floor, throw the ball underhand. Repeat task, throwing ball overhead.

Item	Score					Asst. Device	Arm		Analysis of Joint Motion and Position					
							R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Reach ball on floor	0	1	2	3	4				-	-	Flexion/neutral/ extension	Supination/ neutral/pronation	Flexion/neutral /extension	Extension/neutral/ flexion
throw ball underhand (winding/extend back at shoulder)	0	1	2	3	4				-	-	Flexion/neutral/ extension	Supination/ neutral/pronation	Flexion/neutral /extension	Flexion/neutral/ extension
throw ball underhand (release at shoulder)	0	1	2	3	4						Flexion/neutral/ extension	Supination/ neutral/pronation	Flexion/neutral /extension	Extension/neutral/ flexion
throw ball overhand	0	1	2	3	4				-	-	Flexion/neutral/ extension	Pronation/neutral/ supination	Flexion/neutral /extension	Extension/neutral/ flexion

Remember that for the Analysis of Joint Motion and Position, the scoring options represent less to more function/optimal position for the item in question.

Please select which joint(s) should be **targeted** in the scoring sheet for the specified task.

	Thumb Score		Fingers Score		Wrist Score		Forearm Score		Elbow Score		Shoulder Score	
	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No
Reach ball on floor	○	○	○	○	○	○	○	○	○	○	○	○
Throw ball underhand (winding/extend at shoulder)	○	○	○	○	○	○	○	○	○	○	○	○
Throw ball underhand (release at shoulder)	○	○	○	○	○	○	○	○	○	○	○	○
Throw ball overhead	○	○	○	○	○	○	○	○	○	○	○	○

Other thoughts or notes regarding Task #7: Reach for a medium-sized (e.g., basket ball) ball placed on the floor, throw the ball underhand. Repeat task, throwing ball overhead.

Block 8

Do you have any comments regarding the following task:

Task #8: Put on a T-shirt overhead and take off the T-shirt.

Put on a T-shirt overhead

Take off the T-shirt

For the following task, we would like you to comment if an additional joint should be analyzed. We believe this task is focused more on the ***Elbow and Shoulder***.

Task #8: Put on a T-shirt overhead and take off the T-shirt.

Item	Score					Asst. Device	Arm		Analysis of Joint Motion and Position					
							R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
put on shirt overhead	0	1	2	3	4				-	-	-	Pronated/supinated/neutral	Flexion/neutral/extension	no shoulder movement/partial shoulder movement/full shoulder movement
take off shirt	0	1	2	3	4				-	-	-	Pronated/supinated/neutral	Flexion/neutral/extension	no shoulder movement/partial shoulder movement/full shoulder movement

Remember that for the Analysis of Joint Motion and Position, the scoring options represent less to more function/optimal position for the item in question.

Please select which joint(s) should be **targeted** in the scoring sheet for the specified task.

	Thumb Score		Fingers Score		Wrist Score		Forearm Score		Elbow Score		Shoulder Score	
	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No
Put on a T-shirt overhead	○	○	○	○	○	○	○	○	○	○	○	○
Take off the T-shirt	○	○	○	○	○	○	○	○	○	○	○	○

Other thoughts or notes regarding Task #8: Put on a T-shirt overhead and take off the T-shirt.

Block 9

Do you have any comments regarding the following task:

Task #9: Put on vest/sweater with zipper, fasten the zipper, pull it all the way up, and pull it back down. [Therapist to place zipper either at top or bottom of clothing depending on if the child can complete the task]

Put on vest/sweater with zipper ☐

Fasten the zipper

Pull it all the way up

Pull it back down

Which word do you prefer?

- ○ vest
- ○ sweater

For the following task, we would like you to comment if an additional joint should be analyzed. We believe this task is focused more on the ***Elbow and Shoulder***.

Task #9: Put on ${q://QID51/ChoiceGroup/SelectedChoices} with zipper, fasten the zipper, pull it all the way up, and pull it back down.

Item	Score					Asst. Device	Arm		Analysis of Joint Motion and Position					
							R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
put on	0	1	2	3	4				-	-	-	-	No elbow movement/partial elbow movement/ full elbow movement	No shoulder movement/partial shoulder movement/ full shoulder movement
stabilize zipper	0	1	2	3	4				palm/close/open	No finger movement/partial finger movement/ full finger movement	flexion/extension/ neutral	supination/pronation/neutral	flexion/extension/neutral	External rotation/internal rotation/neutral
Attach zipper	0	1	2	3	4				palm/close/open	No finger movement/partial finger movement/ full finger movement	flexion/extension/ neutral	supination/pronation/neutral	flexion/extension/neutral	External rotation/internal rotation/neutral
pull zipper up to top	0	1	2	3	4				palm/close/open	Extension/neutral/flexion	extension/flexion/ neutral	supination/pronation/neutral	extension/neutral/flexion	Internal rotation/External rotation/neutral
pull zipper all the way down	0	1	2	3	4				palm/close/open	Extension/neutral/flexion	extension/flexion/ neutral	supination/pronation/neutral	flexion/neutral/extension	Internal rotation/external rotation/neutral

Remember that for the Analysis of Joint Motion and Position, the scoring options represent less to more function/optimal position for the item in question.

Please select which joint(s) should be **targeted** in the scoring sheet for the specified task.

	Thumb Score		Fingers Score		Wrist Score		Forearm Score		Elbow Score		Shoulder Score	
	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No
Put on	○	○	○	○	○	○	○	○	○	○	○	○
Stabilize the zipper	○	○	○	○	○	○	○	○	○	○	○	○
Attach the zipper	○	○	○	○	○	○	○	○	○	○	○	○
Pull the zipper all the way up	○	○	○	○	○	○	○	○	○	○	○	○

	Thumb Score		Fingers Score		Wrist Score		Forearm Score		Elbow Score		Shoulder Score	
	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No
Pull the zipper down	○	○	○	○	○	○	○	○	○	○	○	○

Other thoughts or notes regarding Task #9: Put on ${q://QID51/ChoiceGroup/SelectedChoices} with zipper, fasten the zipper, pull it all the way up, and pull it back down. [Therapist to place zipper either at top or bottom of clothing depending on if the child can complete the task]

Block 10

Do you have any comments regarding the following task:

Task #10: Pull down your pants, reach bum/buttocks area, place a sticker on bum/buttocks area [proxy for wiping after bowel movement], place sticker in between legs [proxy for wiping after urination], and pull pants back up. [Therapist to provide loose fitting pants].

Pull down your pants

Reach bum/buttocks area

Place a sticker on bum/buttocks area

Place sticker in between legs

Pull pants back up

Which word do you prefer?

- ○ bum
- ○ buttock

For the following task, we would like you to comment if an additional joint should be analyzed. We believe this task is focused more on the ***Thumb, Wrist, Elbow***.

Task #10: Pull down your pants, reach ${q://QID56/ChoiceGroup/SelectedChoices} area, place a sticker on ${q://QID56/ChoiceGroup/SelectedChoices} area, place sticker in between legs, and pull pants back up.

Item	Score					Asst. Device	Arm		Analysis of Joint Motion and Position					
							R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Pull down	0	1	2	3	4				palm/close/open	-	flexion/neutral/extension	-	no elbow movement/partial elbow movement/full elbow movement	-
Reach bum	0	1	2	3	4				-	-	-	-	flexion/neutral/extension	no shoulder movement/partial shoulder movement/full shoulder movement
Place sticker on bum	0	1	2	3	4				palm/close/open	no finger movement/partial finger movement/full finger movement	extension/neutral/flexion	pronation/neutral/supination	-	-
Place sticker between legs	0	1	2	3	4				palm/close/open	no finger movement/partial finger movement/full finger movement	extension/neutral/flexion	supination/neutral/pronation	-	-
Pull up pants	0	1	2	3	4				palm/close/open	no finger movement/partial finger movement/full finger movement	-	-	no elbow movement/partial elbow movement/full elbow movement	-

Remember that for the Analysis of Joint Motion and Position, the scoring options represent less to more function/optimal position for the item in question.

Please select which joint(s) should be **targeted** in the scoring sheet for the specified task.

	Thumb Score		Fingers Score		Wrist Score		Forearm Score		Elbow Score		Shoulder Score	
	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No
Pull down	○	○	○	○	○	○	○	○	○	○	○	○
Reach bum	○	○	○	○	○	○	○	○	○	○	○	○
Place sticker on bum	○	○	○	○	○	○	○	○	○	○	○	○
Place sticker in between legs	○	○	○	○	○	○	○	○	○	○	○	○
Pull up pants	○	○	○	○	○	○	○	○	○	○	○	○

Other thoughts or notes regarding Task #10: Pull down your pants, reach ${q://QID56/ChoiceGroup/SelectedChoices} area, place a sticker on ${q://QID56/ChoiceGroup/SelectedChoices} area, place sticker in between legs, and pull pants back up.

Block 11

Do you have any comments regarding the following task:

Task #11: Put on a sock and take it off. [Note: Do not score this task if the child has a lower extremity contracture that prohibits task completion].

Put on a sock

Take it off

For the following task, we would like you to comment if an additional joint should be analyzed. We believe this task is focused more on the ***Thumb and Elbow***.

Task #11: Put on a sock and take it off.

Item	Score					Asst. Device	Arm		Analysis of Joint Motion and Position					
							R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Put on sock over toe	0	1	2	3	4				palm/close/open	Extension/neutral/flexion	-	-	Flexion/neutral/extension	-
Pull sock over heel	0	1	2	3	4				palm/close/open	Extension/neutral/flexion	-	-	extension/neutral/flexion	-

Remember that for the Analysis of Joint Motion and Position, the scoring options represent less to more function/optimal position for the item in question.

Please select which joint(s) should be **targeted** in the scoring sheet for the specified task.

	Thumb Score		Fingers Score		Wrist Score		Forearm Score		Elbow Score		Shoulder Score	
	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No
Put on sock over toe	○	○	○	○	○	○	○	○	○	○	○	○
Pull sock over heel	○	○	○	○	○	○	○	○	○	○	○	○

Other thoughts or notes regarding Task #11: Put on a sock and take it off. [Note: Do not score this task if the child has a lower extremity contracture that prohibits task

completion].

Block 12

Do you have any comments regarding the following task:

Task #12: Show us how you move from lying down on your back to a sitting position

Lying to sitting

For the following task, we would like you to comment if an additional joint should be analyzed. We believe this task is focused more on the ***Wrist, Forearm, Elbow***.

Task #12: Show us how you move from lying down on your back to a sitting position

Item	Score					Asst. Device	Arm		Analysis of Joint Motion and Position					
							R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Lying to sitting	0	1	2	3	4				-	-	flexion/neutral/extension	supination/neutral/pronation	flexion/neutral/extension	-

Remember that for the Analysis of Joint Motion and Position, the scoring options represent less to more function/optimal position for the item in question.

Please select which joint(s) should be **targeted** in the scoring sheet for the specified task.

	Thumb Score		Fingers Score		Wrist Score		Forearm Score		Elbow Score		Shoulder Score	
	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No
Lying to sitting	○	○	○	○	○	○	○	○	○	○	○	○

Other thoughts or notes regarding Task #12: Show us how you move from lying down on your back to a sitting position

Block 13 - Qualitative Questions

In addition to the quantitative tasks and scoring above, we'd like to ask a few descriptive questions.
Please provide comments and feedback on the next 3 qualitative questions.

Does the child use their arms for:

- Using a mobility device ☐ Yes ☐ No. If yes, describe:

Does the child use their arms for:

- Shifting /changing/moving your body (e.g., getting on or off couch/toilet/etc.) ☐ Yes ☐ No. If yes, describe:

Are you using a splint for the tasks included: ☐ Yes ☐ No; ☐ Right ☐ Left. If yes, describe:

Block 14 - Overall Comments

Thank you for completing the survey. Please provide any feedback regarding the layout of the scoring sheet, the overall content of the SHAPE-UP, if any items/tasks are missing, or any other comments.

Appendix 5. Manuscript 2 – Qualtrics Clinician Survey Round 3

Block 0 - Introduction

Thank you for participating in the third and final round of the SHAPE-UP Delphi. We have taken into account all your comments and feedback and have modified the SHAPE-UP outcome measure. The yellow highlights indicate all the modifications and edits made to the SHAPE-UP between Round 2 and Round 3.

Please note there are now 11 tasks as well as 3 descriptive questions. Each task is further broken down into a series of items and scored accordingly. Some tasks are unilateral, others bilateral, but this has been accounted for in the scoring table.

As a reminder, the purpose of the SHAPE-UP is to assess upper extremity (UE) function in children with AMC with upper limb involvement. The SHAPE-UP will describe the impairments, activity limitations, and participation restrictions in the performance of daily tasks in children with AMC to guide treatment decision-making and evaluation treatment effectiveness for the UE.

The scoring guide below describes how each item within the 11 tasks will be scored.

Task Completion
0 = **unable** - The child is unable to complete any component of the task.
1 = **partial** completion of task **passively** - The child can partially complete the task using passive range of motion.
2 = **partial** completion of task **actively** - The child can partially complete the task using active range of motion.

3 = **completion** of task **passively** - The child can complete the task using passive range of motion.
4 = **completion** of task **actively** - The child can complete the task using active range of motion.
Analysis of Joint Motion and Position
Each joint of the UE (**Thumb, Fingers, Wrist, Forearm, Elbow, Shoulder**) is labeled and categorized from **least to most optimal position** for the specific item being assessed. If the item scoring option is "-", the focus of the task and item is not on that specific joint.

In addition, we strongly recommend you download the following PDF as a reference guide for the scoring tables for all 11 tasks and 3 descriptive questions. This will allow you to get a complete picture of the SHAPE-UP in it's entirety.

For this final round Delphi, we ask that you look at the SHAPE-UP as a whole and provide any comments regarding scoring, overall layout, and clarity of content.

We appreciate your time and efforts in completing this survey.

Block 1

Task #1. Pick up a cheerio, bring it to your mouth, place it back down in front of you, and let it go.

Item	Task completion					Arm		Analysis of Joint Motion and Position					
						R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Pick up cheerio	0	1	2	3	4			Palm/Close/Open	Flexion/Neutral/Extension	Flexion/Neutral/Extension	-	-	-
Bring it to mouth	0	1	2	3	4			-	-	-	Pronated/ /Neutral/Supination	Extension/Neutral/Flexion	No External Rotation/Partial External Rotation/Full External Rotation
Place it back down in front of you	0	1	2	3	4			-	-	-	Supination/Neutral/ Pronation	Flexion/Neutral/Extension	-
Let it go	0	1	2	3	4			Palm/Close/Open	Flexion/Neutral/Extension	Flexion/Neutral/Extension	-	-	-

Remember that for the Analysis of Joint Motion and Position, the scoring options indicate less to more function/optimal position for the specific item within the task.

Additional comments regarding Task #1: Pick up a cheerio, bring it to your mouth, place it back down in front of you, and let it go.

Block 2

Task #2: Grasp a water bottle, bring it to your mouth, place it back down in front of you, and let it go.

Item	Score					Arm		Analysis of Joint Motion and Position					
						R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Grasp a water bottle	0	1	2	3	4			Palm/Close/Open	Flexion/Neutral/Extension	Flexion/Neutral/Extension	Supination/Neutral/Pronation	-	-
Bring it to mouth	0	1	2	3	4			-	-	Flexion/Neutral/Extension	Pronation/ /Neutral/Supination	Extension/Neutral/Flexion	No External Rotation/Partial External Rotation/Full External Rotation
Place it back down in front of you	0	1	2	3	4			-	-	Flexion/Neutral/Extension	Supination/Neutral/ Pronation	Flexion/Neutral/Extension	-
Let it go	0	1	2	3	4			Palm/Close/Open	Flexion/Neutral/Extension	Flexion/Neutral/Extension	Supination/Neutral/Pronation	-	-

Remember that for the Analysis of Joint Motion and Position, the scoring

options indicate less to more function/optimal position for the specific item within the task.

Additional comments regarding Task #2. Grasp a water bottle, bring it to your mouth, place it back down in front of you, and let it go.

Block 3

Task #3. Open the jar, pour out a few cubes, string 3 together, and close the jar.

Item	Score					Arm		Analysis of Joint Motion and Position					
						R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Stabilizes jar	0	1	2	3	4			Palm/Close/Open	No Finger Movement/Partial Finger Movement/ Full Finger Movement	Extension/ Flexion/Neutral	Supination/Pronation/ Neutral	-	-
Open jar	0	1	2	3	4			Palm/Close/Open	No Finger Movement/Partial Finger Movement/ Full Finger Movement	Extension/ Neutral/ Flexion	Supination/Neutral/ Pronation	-	-
Pour out	0	1	2	3	4			Palm/Close/Open	No Finger Movement/Partial Finger Movement/ Full Finger Movement	-	Supination/Neutral/ Pronation	-	-
Hold bead	0	1	2	3	4			Palm/Close/Open	Fingers flexed (hand fisted) /Hand opened (not fisted)/Grasp between thumb and fingers (opposition)	-	-	-	-
String 3	0	1	2	3	4			Palm/Close/Open	Fingers flexed (hand fisted) /Hand opened (not fisted)/Grasp between thumb and fingers (opposition)	-	-	-	-
Close jar	0	1	2	3	4			Palm/Close/Open	No Finger Movement/Partial Finger Movement/ Full Finger Movement	Extension/ Neutral/ Flexion	Supination/Neutral/ Pronation	-	-

Remember that for the Analysis of Joint Motion and Position, the scoring options indicate less to more function/optimal position for the specific item within the task.

Additional comments regarding Task #3: Open the jar, pour out a few cubes, string 3 together, and close the jar.

Block 4

Task #4. Pick up the marker, write your name on this piece of paper with marker, fold the paper, and cut it using the scissors.

[Therapist to place uncapped marker across the midline]

Item	Score					Arm		Analysis of Joint Motion and Position					
						R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Pick up the marker	0	1	2	3	4			Palm/Close/Open	No Finger Movement/Partial Finger Movement/ Full Finger Movement	Flexion/Neutral/ Extension	Supination/Neutral/Pronation	-	-
Write name	0	1	2	3	4			Palm/Close/Open	No Finger Movement/Partial Finger Movement/ Full Finger Movement	Flexion/Neutral/ Extension	Supination/Neutral/Pronation	-	-
Fold paper	0	1	2	3	4			Palm/Close/Open	No Finger Movement/Partial Finger Movement/ Full Finger Movement	Flexion/Neutral/ Extension	Supination/Neutral/Pronation	-	-
Cut paper	0	1	2	3	4			Palm/Close/Open	No Finger Movement/Partial Finger Movement/ Full Finger Movement	Flexion/Neutral/ Extension	Supination/Pronation/Neutral	-	-
Stabilize paper to cut	0	1	2	3	4			Palm/Close/Open	No Finger Movement/Partial Finger Movement/ Full Finger Movement	Flexion/Neutral/ Extension	Supination/Pronation/Neutral	-	-

Remember that for the Analysis of Joint Motion and Position, the scoring options indicate less to more function/optimal position for the specific item within the task.

Additional comments regarding Task #4. Pick up the marker, write your name on this piece of paper with marker, fold the paper, and cut it using the scissors.

Block 5

Task #5: Pick up the Play-Doh using the fork and bring to mouth.

[Therapist places piece of Play-Doh in front of child]

Item	Score					Arm		Analysis of Joint Motion and Position				
						R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Pick Up the Play-Doh	0	1	2	3	4			Palm/Close/Open	Fingers flexed (hand fisted) /Hand opened (not fisted)/Grasp between thumb and fingers (opposition)	Flexion/Extension/Neutral	Supination/Pronation/ Neutral	-	No External Rotation/ Partial External Rotation/ Full External Rotation
Bring to Mouth	0	1	2	3	4			-	-	-	Pronation//Neutral/Supination	Extension/Neutral/Flexion	No External Rotation/ Partial External Rotation/ Full External Rotation

Remember that for the Analysis of Joint Motion and Position, the scoring options indicate less to more function/optimal position for the specific item within the task.

Additional comments regarding Task #5: Pick up the Play-Doh using the fork and bring to mouth.

Block 6

Task #6: Grasp a small-size ball (e.g., tennis ball), throw the ball underhand. Repeat task, throwing ball overhead.

Item	Score					Arm		Analysis of Joint Motion and Position					
						R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Grasp small size ball	0	1	2	3	4			-	-	Flexion/Neutral/ Extension	Supination/ Neutral/Pronation	Flexion/Neutral /Extension	Extension/Neutral/ Flexion
Wind up in preparation to throw ball underhand	0	1	2	3	4			-	-	Flexion/Neutral/ Extension	Supination/ Neutral/Pronation	Flexion/Neutral /Extension	Flexion/Neutral/ Extension
Throw ball underhand	0	1	2	3	4			-	-	Flexion/Neutral/ Extension	Supination/ Neutral/Pronation	Flexion/Neutral /Extension	Extension/Neutral/ Flexion
Wind up in preparation to throw ball overhead	0	1	2	3	4			-	-	Flexion/Neutral/ Extension	Supination/ Neutral/Pronation	Flexion/Neutral /Extension	Flexion/Neutral/ Extension
Throw ball overhead								-	-	Flexion/Neutral/ Extension	Supination/ Neutral/Pronation	Flexion/Neutral /Extension	Extension/Neutral/ Flexion

Remember that for the Analysis of Joint Motion and Position, the scoring options indicate less to more function/optimal position for the specific item within the task.

Additional comments regarding Task #6: Grasp a small-size ball (e.g., tennis ball), throw the ball underhand. Repeat task, throwing ball overhead.

Block 7

Task #7: Grasp a medium-size ball (e.g., basket ball), throw the ball underhand. Repeat task, throwing ball overhead.

Underhand **Overhead**

Grasping bilaterally ☐ Yes ☐ No Grasping bilaterally ☐ Yes ☐ No

Throwing bilaterally ☐ Yes ☐ No Throwing bilaterally ☐ Yes ☐ No

Item	Score					Arm		Analysis of Joint Motion and Position					
						R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Grasp medium size ball	0	1	2	3	4			-	-	Flexion/Neutral/ Extension	Supination/ Neutral/Pronation	Flexion/Neutral /Extension	Extension/Neutral/ Flexion
Wind up in preparation to throw ball underhand	0	1	2	3	4			-	-	Flexion/Neutral/ Extension	Supination/ Neutral/Pronation	Flexion/Neutral /Extension	Flexion/Neutral/ Extension
Throw ball underhand	0	1	2	3	4			-	-	Flexion/Neutral/ Extension	Supination/ Neutral/Pronation	Flexion/Neutral /Extension	Extension/Neutral/ Flexion
Wind up in preparation to throw ball overhead								-	-	Flexion/Neutral/ Extension	Supination/ Neutral/Pronation	Flexion/Neutral /Extension	Extension/Neutral/ Flexion
Throw ball overhead	0	1	2	3	4			-	-	Flexion/Neutral/ Extension	Supination/Neutral/ Pronation	Flexion/Neutral /Extension	Extension/Neutral/ Flexion

Remember that for the Analysis of Joint Motion and Position, the scoring options indicate less to more function/optimal position for the specific item within the task.

Additional comments regarding Task #7: Grasp a medium-size ball (e.g., basket ball), throw the ball underhand. Repeat task, throwing ball overhead.

Block 8

Task #8: Put on and take off a T-shirt.

[Note that the following items do not need to be completed in the presented order.]

Item	Score					Arm		Analysis of Joint Motion and Position					
						R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Put T-shirt over head	0	1	2	3	4			-	-	-	Pronated/Supinated/Neutral	Flexion/Neutral/Extension	No Shoulder Movement/Partial Shoulder Movement/Full Shoulder Movement
Put arms in sleeves	0	1	2	3	4			-	-	-	Pronated/ Neutral/Supinated	Flexion/Neutral/Extension	Internal rotation/neutral/External rotation
Pull down T-shirt	0	1	2	3	4			Palm/Close/Open	Fingers flexed (hand fisted) /Hand opened (not fisted)/Grasp between thumb and fingers (opposition)	Extension/Flexion/ Neutral	Pronated/Supinated/Neutral	Flexion/Neutral/Extension	No Shoulder Movement/Partial Shoulder Movement/Full Shoulder Movement
Remove arms from sleeves	0	1	2	3	4			-	-	-	Pronated/Supinated/Neutral	Flexion/Neutral/Extension	No Shoulder Movement/Partial Shoulder Movement/Full Shoulder Movement
Remove T-shirt	0	1	2	3	4			Palm/Close/Open	No finger movement/partial finger movement/full finger movement	Flexion/neutral/ extension	Pronated/Supinated/Neutral	Flexion/Neutral/Extension	Flexion/neutral/ extension/

Remember that for the Analysis of Joint Motion and Position, the scoring options indicate less to more function/optimal position for the specific item within the task.

Additional comments regarding Task #8: Put on and take off a T-shirt.

Block 9

Task #9: Put on zippered item on upper body, fasten the zipper, pull it all the way up, and pull it back down.

[Therapist to place zipper either at top or bottom depending on if the child can complete the task; if child cannot fasten the zipper, therapist to attach zipper for them.]

Item	Score					Arm		Analysis of Joint Motion and Position					
						R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Put on item of clothing	0	1	2	3	4			-	-	-	-	No Elbow Movement/ Partial Elbow Movement/ Full Elbow Movement	No Shoulder Movement/ Partial Shoulder Movement/ Full Shoulder Movement
Stabilize zipper	0	1	2	3	4			Palm/Close/Open	No Finger Movement/ Partial Finger Movement/ Full Finger Movement	Flexion/ Extension/ Neutral	Supination/ Pronation/ Neutral	Flexion/Extension/Neutral	External Rotation/ Internal Rotation/ Neutral
Fasten zipper	0	1	2	3	4			Palm/Close/Open	No Finger Movement/ Partial Finger Movement/ Full Finger Movement	Flexion/ Extension/ Neutral	Supination/ Pronation/ Neutral	Flexion/Extension/Neutral	External Rotation/ Internal Rotation/ Neutral
Pull zipper all the way up	0	1	2	3	4			Palm/Close/Open	Extension/Neutral/Flexion	Extension/ Flexion/ Neutral	Supination/ Pronation/ Neutral	Extension/Neutral/Flexion	Internal Rotation/ External Rotation/ Neutral
Pull zipper all the way down	0	1	2	3	4			Palm/Close/Open	Extension/Neutral/Flexion	Extension/ Flexion/ Neutral	Supination/ Pronation/ Neutral	Flexion/Neutral/Extension	Internal Rotation/ External Rotation/ Neutral

Remember that for the Analysis of Joint Motion and Position, the scoring options indicate less to more function/optimal position for the specific item within the task.

Additional comments regarding Task #9: Put on zippered item on upper body, fasten the zipper, pull it all the way up, and pull it back down.

Block 10

Task #10: Pull down your pants, reach buttocks area from the back while holding 2 squares of toilet paper, and now reach between legs from the front while holding 2 squares of toilet paper, and pull pants back up.

[Therapist to provide loose fitting pants]

Item	Score					Arm		Analysis of Joint Motion and Position					
						R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Pull down pants	0	1	2	3	4			Palm/Close/Open	-	Flexion/Neutral/ Extension	-	No Elbow Movement/ Partial Elbow Movement/ Full Elbow Movement	-
Reach buttocks area [back]	0	1	2	3	4			-	-	-	-	Flexion/Neutral/Extension	No Shoulder Movement/ Partial Shoulder Movement/ Full Shoulder Movement
Reach between legs [front]	0	1	2	3	4			Palm/Close/Open	No Finger Movement/ Partial Finger Movement/ Full Finger Movement	Extension/Neutral/ Flexion	Supination/Neutral/ Pronation	-	-
Pull up pants	0	1	2	3	4			Palm/Close/Open	No Finger Movement/ Partial Finger Movement/ Full Finger Movement	-	-	No Elbow Movement/ Partial Elbow Movement/ Full Elbow Movement	-

Remember that for the Analysis of Joint Motion and Position, the scoring options indicate less to more function/optimal position for the specific item within the task.

Additional comments regarding Task #10: Pull down your pants, reach buttocks area from the back while holding 2 squares of toilet paper, and now reach between legs from the front while holding 2 squares of toilet paper, and pull pants back up.

Block 11

Task #11: Put on a sock and take it off.
[If LE contracture prohibits the task, check □ NO SCORE]

Item	Score					Arm		Analysis of Joint Motion and Position					
						R	L	Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Put on sock over toes	0	1	2	3	4			Palm/Close/Open	Extension/Neutral/Flexion	-	-	Flexion/Neutral/Extension	-
Pull sock over heel	0	1	2	3	4			Palm/Close/Open	Extension/Neutral/Flexion	-	-	Extension/Neutral/Flexion	-

Remember that for the Analysis of Joint Motion and Position, the scoring options indicate less to more function/optimal position for the specific item within the task.

Additional comments regarding Task #11: Put on a sock and take it off.

Block 12 - Descriptive Questions

Does the child use their arms to:

- Propel a wheelchair ☐Yes ☐ No. If yes, describe: _____

- Use a walking aid (cane, crutch, walker) ☐Yes ☐ No. If yes, describe: _____

-Other: _____ ☐Yes ☐ No. If yes, describe: _____

Any additional comments regarding Descriptive Question #1 above:

Block 13

Does the child use their arms to perform the following transfers:

Transfers	Uses their arms		Cannot use their arms	Cannot complete due to LE contracture	Describe
	Right	Left			
Bed positioning					
Lying to sitting					
Sitting to standing					
Toilet transfer					
Bathtub/shower transfer					
Getting in and out of a car					
Other:					
Other:					
Other:					

Any additional comments regarding Descriptive Question #2 above:

Block 14

Does the child use a splint/orthosis when performing the tasks included in the SHAPE-UP:

☐ Yes ☐ No. If yes indicate side: ☐ Right ☐ Left, and describe type of splint and its use: _____

Any additional comments regarding Descriptive Question #3 above:

Block 14 - Overall Comments

Please provide any overall feedback regarding the layout of the scoring sheet, the overall content of the SHAPE-UP, or any other comments.

Appendix 6. SHAPE-UP Recruitment Flyer

20215864
#32458133.0
IRB Approved at the
Protocol Level
Nov 19, 2021

Shriners Hospital Arthrogryposis Pediatric Evaluation – UPper Extremity

SHAPE-UP

We are recruiting participants for a research study involving the upper extremity function of children and adolescents with arthrogryposis.

What is the purpose of this study?
The research study aims to develop an outcome measure specific to the **upper extremity function** of children and adolescents with arthrogryposis multiplex congenita (AMC).

Who can participate?
Children and adolescents
- with multiple congenital contractures (arthrogryposis)
- between the ages of 1 and 21 years

Youth and parents who understand and communicate in English or [Language]

What will you be asked to do?
Youth who agree to participate will be asked to perform a set of tasks using their arms, hands, and fingers.

Who can I contact for additional information?
To participate or if you have any questions please contact [Name] at [Contact email address]. or at [Contact #].

Who are investigators on this study?
Dr. Noémi Dahan-Oliel is the principal investigator: ndahan@shrinenet.org
[Site Investigator's Name] is the site investigator: [Contact email address]

This study is funded by a Shriners Developmental Grant (2021-2023)

Hôpitaux Shriners pour enfants® Shriners Hospitals for Children® Canada

Appendix 7. SHAPE-UP Letter of Invitation to Participate in a Research Study

Shriners Hospitals for Children®

WRIST THUMB SHAPE-UP AND ELBOW

Shriners Hospitals for Children - Canada
1003, boulevard Décarie
Montréal, Québec H4A 0A9

LETTER OF INVITATION TO PARTICIPATE IN A RESEARCH STUDY

Measure for the upper extremity in arthrogryposis: Development and psychometrics

Noémi Dahan-Oliel, PhD OT
Shriners Hospitals for Children - Canada
1003, boulevard Décarie
Montréal, Québec H4A 0A9

You are being invited to participate in a voluntary research study. You are being contacted because you or your child has arthrogryposis, and is being treated at Shriners Children's – Canada. The study includes five other Shriners Children's sites across North America. The research study aims to develop an outcome measure specific to the upper extremity function of children and adolescents with arthrogryposis. The research study includes a set of tasks performed using arms, hands, and fingers. The findings from this project will help us develop an upper extremity assessment specific to children with arthrogryposis, in order to better understand the needs of children with arthrogryposis and evaluate the effects of surgery to the upper extremity.

If you agree to participate in this study, we will schedule a clinic visit at Shriners Children's - Canada. During this visit, we will ask you or your child to perform a set of tasks using upper extremities; and we will ask questions about upper extremity function and past surgery, splinting or therapy that you or your child may have had, and/or is currently having.

Any information you provide will be used for research purposes only and your decision to participate or not in this study will not affect the services you or your child receives at the Shriners Children's in any way.

If you are interested in knowing more about this study or would like to participate, please contact Sena Tavukcu our study coordinator by email at SeyhanSena.Tavukcu@shrinenet.org or by phone at 514-842-4464 ext.2310

Looking forward to hearing back from you!

v.22AUG2022

Appendix 8. SHAPE-UP Consent Form Manuscript 3

Shriners Hospitals for Children- Canada

INFORMED CONSENT TO TAKE PART IN A RESEARCH STUDY
FOR: PARENTS OF CHILDREN 1-13 YEARS, or YOUTH UNABLE TO CONSENT, or YOUTH 14-21 YEARS

TITLE:	**Measure for the upper extremity in arthrogryposis: Development and psychometrics**
PROTOCOL NO.:	**CAN2103**
FUNDING SPONSOR:	Shriners Hospitals for Children, Grant #79152
REGULATORY SPONSOR:	Shriners Hospitals for Children, International Headquarters 2900 Rocky Point Drive Tampa, Florida 33607
PRINCIPAL INVESTIGATOR:	Noemi Dahan-Oliel, PhD, OT Shriners Hospital for Children- Canada 1003 Boul. Décarie, Montreal, Quebec
CO-INVESTIGATOR:	Chantal Janelle, MD Shriners Hospital for Children- Canada
STUDY COORDINATOR:	Kathleen Montpetit, MSc OT 514-842-4464 x 2263 kmontpetit@shrinenet.org Seyhan Sena Tavukcu, BSc 514-842-4464 x 2310 SeyhanSena.Tavukcu@shrinenet.org

PARTICIPANT: _____

When we say "you" in this consent form, we mean you or your child; "we" means the doctors and other staff.

SUMMARY

You are being asked to take part in a research study. The purpose of this consent form is to help you decide if you want to be in the research study.

Research studies include only people who want to take part. Before you decide if you want to take part, it is important that you read and understand this consent form. Please take your time to make your decision. Discuss it with your friends and family. We encourage you to include your child in the discussion and decision if she or he is able to understand. Please ask questions about anything that you do not understand before deciding whether or not to participate.

The person in charge of this study, locally, is Dr. Noémi Dahan-Oliel. There may be other people on the research team helping during the study.

Things to know and understand before deciding to take part in a research study:

- The main goal of a research study is to learn things to help patients in the future.
- The main goal of regular medical care is to help each patient.
- The decision to join or not join the research study will not cause you to lose any medical benefits. If you decide not to take part in this study, your doctor will continue to treat you.
- Parts of this study may involve standard medical care. Standard care is the treatment normally given for a certain condition or illness.
- After reading the consent form and having a discussion with the research staff, you should know which parts of the study are experimental and which are standard medical care.

Why are you being asked to take part in this study?

You are being asked to take part in a research study because you are a caregiver or parent of a child between the ages of 1 to 13 years, or a youth between 14 and 21 years of age with a clinical diagnosis of arthrogryposis multiplex congenita (AMC) attending Shriners Hospital for Children – Canada.

The purpose of this study is to develop an outcome measure called Shriners Hospital Arthrogryposis Pediatric Evaluation-UPper extremity (SHAPE-UP) that will measure how you complete simple daily task using your arms and hands. The SHAPE-UP will help us understand the way in which tasks are completed. The SHAPE-UP will help healthcare professionals determine a reliable way to measure the function of your arms and hands.

We hope to:

- Overall, develop an upper extremity assessment specific to children with AMC
- Determine if healthcare professionals find the SHAPE-UP is appropriate and easy to use
- Determine that the scoring is consistent for all healthcare professionals and that we are measuring the right thing

How many people will take part in the study?

About 120 children/youthwill take part in this study. At this site, about 20 children will take part in this study.

What will happen on this study?

If you agree to be in this study, then you will be asked to do the following tests and/or procedures at the designated time points.

Before you begin the study…

- The clinical research coordinator (CRC) or principal investigator (PI) will briefly explain the study's objectives.
- Your informed consent will be obtained by the PI or CRC.

During the study with your consent…

- You will be given the option to complete the assessment at the hospital after your clinic visit or at home with videoconferencing.
- you will be asked questions about your medical history and current level of functioning. We may also extract this information from your medical chart.

- You will be asked to complete a series of tasks (ex: opening a jar, zipping a jacket, bringing fork to mouth) specific to your age and expected level of functioning during a hospital visit or during a remote session.
- You will be recorded while completing these tasks in order to gain a better understanding of how you perform these activities.
- If you agree the recording of you completing the simple tasks may be shown to other participating members of the research team at the other participating Shriners Hospitals for Children to establish the properties of this new measure as well the recording may be used for training purposes.
- In order to verify the reliability of the SHAPE-UP, you may be asked to complete the same series of tasks a second time at a later date (between 1 and 4 weeks after your first assessment).
  If you are scheduled to undergo a surgery to your upper extremity, you may be asked to complete the same series of tasks up to three times: before the surgery, 4-6 months after surgery, and one year after surgery in order to determine if the SHAPE-UP can measure changes resulting from the surgery.

When you are finished:

- The information collected will allow us to develop an assessment that measures the function of the arms and hands of children with AMC and to determine if the SHAPE-UP is easy to use and evaluating the appropriate things.

What will happen in this study that is "research"?
There are no experimental tests used in this study. You will participate in completing tasks specific to your age.

How long does the study last?
Your participation in the study session will last approximately 45 minutes, the time to complete age-appropriate tasks. You may be invited to complete additional sessions.
This study is expected to last approximately three years.

Can you stop being in the study?
Yes. You can drop out of the study at any time and no one will be upset. It will not affect your other care received at Shriners Hospitals for Children. You can do this by contacting Dr. Noemi Dahan-Oliel at 514-842-4464 extension 2278 or by email at ndahan@shrinenet.org. It is important to know that there will be no negative impact if you choose to stop.

If you drop out of the study, no new health information identifying you will be gathered after that date. Information that has already been gathered may still be used and given to others.

The study investigator may decide to take you off this study without your consent under the following circumstances:

- If she or he believes that it is in your best interest
- If study procedures are not followed or if visits are not kept

What are the risks of the study?
This study involves no more than minimal risks, which means that we expect that you will have no more risk than those you have in your normal daily life or routine physical examination or tests. The only risk is the possible loss of confidentiality.
Please let Dr. Noemi Dahan-Oliel at 514-842-4464 x 2278 or the study coordinator know if you experience discomfort during the study-related evaluations.

Are there benefits to taking part in the study?
You will not directly benefit from your participation in this study. Possible benefits that might result from this research are that your involvement will help develop a hand and arm specific assessment to assist in the rehabilitation treatment of children with AMC.

What other options are available to you?
This is not a treatment study. Your alternative is not to be in this study.

What are the costs to taking part in the study?
To offset any inconvenience for the additional time required due to study participation, you will be provided a 20$ gift card.

In the case of injury or illness resulting from this study, medical treatment is available. Please notify the investigator as soon as possible if you need to seek medical care for an injury or illness resulting from this study. Although Shriners Hospitals for Children is not able to offer financial compensation for an injury or illness resulting from this study, to the extent Shriners Hospitals for Children provides the needed medical treatment at its facility, that treatment may be provided at no cost to you. If you obtain that care somewhere else, your usual healthcare coverage may apply. Although Shriners Hospitals for Children has not set aside any funds to pay you for injury or illness, you do not give up any of your legal rights by signing this form.

What are your rights if you take part in this study?
Voluntary Participation: Taking part in this study is your choice. You may choose not to take part in the study or you may leave at any time. Your decision about being in this study or refusal will not affect your care at Shriners Hospitals for Children. If you have any questions about the study, you can always talk to one of the study staff. Do not sign this form unless you have had the chance to ask questions and have received clear answers.

New Findings: We do not expect to find any new information that may affect your health or willingness to stay in this study during your brief involvement. We will tell you if there were any new findings.

Who will be paying for the study?
Shriners Hospitals for Children will pay for this study.

Whom can you call if you have questions or problems?
If you have any questions, please ask us. It is important that you tell the study investigator if you feel that you have been injured because of taking part in this study. In the event of research-related injury, please call Dr. Noemi Dahan-Oliel at 514-842-4464 x 2278 or email at ndahan@shrinenet.org

If you have questions about your rights as a research subject or if you have questions, concerns or complaints about the research, you may contact: Ms. Ilde Lepore, McGill Institutional Review Board, at 514-398-8302 or ilde.lepore@mcgill.ca

AUTHORIZATION TO USE AND DISCLOSE INFORMATION FOR RESEARCH PURPOSES

What information may be used and given to others?
The research team may use your personal and health information (which is de-identified), using only the minimal health information needed to complete study.

Personal health information (PHI) obtained from the following records may be used or disclosed:

- Past medical history as it relates to the research study since diagnosis of AMC given.
- Present medical records as it relates to the research study, including:
  - History and Physical Examination

  - Progress Notes
  - Operative Reports
- Study Questionnaires
- Video recording of the assessment
- Research records
- Records about phone calls made as part of this research
- Records about your study visits

Who may use and give out information about me in connection with this research study?

- The principal investigator, Dr. Noemi Dahan-Oliel at 514-842-4464 x 2278
- Other members of the research team affiliated with Shriners Hospitals for Children who might need to use or disclose your information in connection with this study.

Who might get this information?

The people mentioned above may share or disclose your health information with the following entities or people in connection with this research study. The recipients of this information, while monitoring the research study, may also review and copy your original SHC records.

- Shriners Hospitals for Children
- Representatives from McGill University may use your information in connection with this research study.

How will this information be kept private?

We will do our best to make sure that the personal information in your medical record will be kept private. However personal information such as medical information and names will be replaced by a study number. Videotaping will focus on your arms and hands as much as possible and every effort will be made not to show your face. However, absolute confidentiality cannot be guaranteed because of the need to give information to these parties. Your personal information may also be given out if required by law.

Why will this information be used and/or given to others?

- To do the research in this study,
- To examine results related to this study, and
- To see if the this study was done right

The results of this research study may be presented at meetings or in publications. Your name and other personal information will not be used in those presentations.

What if you decide not to give permission to use your health information?

Then you will not be able to be in this research study.

What if you decide not to give permission to give out your health information?

Then you will not be able to be in this research study.

Can you review or copy your information?

Yes, but only after the research is over.

Can you withdraw or revoke (cancel) your permission to use and give out your health information?

This permission will be good until seven years following the study closure.

You may withdraw or take away your permission to use and disclose your health information at any time. You do this by sending a written notice to the study doctor. If you withdraw your permission, you will not be able to stay in this study.

When you withdraw your permission, no new health information identifying you will be gathered after that date. You may withdraw any Information that has already been gathered. You will be asked specifically if the researchers wish to retain any already collected study data.

Is your health information protected after it has been given to others?
There is a risk that your information may be given to others without your permission. Any information that is shared may no longer be protected by federal privacy rules.

Measure for the upper extremity in arthrogryposis: Development and psychometrics

My signature below will show that I acknowledge and agree that:

- **I have read this consent form (or it has been read to me)**
- **I (my child) will freely take part in this research study,**
- **I have had a chance to ask questions and all of my questions have been answered**
- **I authorize the release of medical and research records for the purpose of this study**
- **I have read and understood the information above**
- **By signing this consent form, I have not given up any of my legal rights.**

You will be given a signed and dated copy of this informed consent form for you to keep.

I agree to have the videorecording shared with the research team to determine the properties of the measure and with clinicians for training purposes:

☐ **Yes** ☐ **No**

I agree to be re-contacted for additional assessment sessions at a later date using the SHAPE-UP measure.

☐ **Yes** ☐ **No**

CONSENT SIGNATURE:

Signature of Parent or Legally Authorized Representative for children under 14 or unable to consent	Date (mm/dd/yyyy)	Name and relationship to participant

OR

Signature of Participant between 14 and 21 years of age	Date (mm/dd/yyyy)

Using language that is understandable and appropriate, I have discussed this project and the items listed above with the participant and/or his authorized representative.

Signature of the person who conducted the informed consent discussion	Date (mm/dd/yyyy)
Print Name	Study Role

I have witnessed the entire informed consent discussion. I attest that the information in the consent form and any other written information was accurately explained to, an apparently understood by, the participant or the participant's legally acceptable representative, and that informed consent was freely given.

Signature of Impartial Witness	Date (mm/dd/yyyy)

(Impartial witness is a person observing or participating in the consent process. This person is independent of the study and cannot be unfairly influenced by the people involved in the study. Regulations only require the use of an impartial witness when either a subject/LAR cannot read and/or does not speak in the language of the approved ICF document. Please refer to the SHC policy on Obtaining Informed Consent-Process and Documentation for more details regarding the responsibilities and requirements of the impartial witness.)

Appendix 9. SHAPE-UP Case Report Form Manuscript 3

Shriners Hospitals for Children®

Study ID:	Date completed (mm/dd/yyyy):

Measure for the upper extremity in arthrogryposis: Development and psychometrics

Demographic information.

Sex: ☐ Male ☐ Female	**Date of Birth (mm/dd/yyyy):**
Type of AMC: ☐ Amyoplasia ☐ Distal arthrogryposis (mainly hand and feet involvement) ☐ Other (central nervous system involvement, chromosomal abnormalities, etc.) ☐ Don't know, describe: _____	

Clinical information. Joint involvement:

i) Jaw	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
ii) Right Shoulder	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
iii) Left shoulder	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
iv) Are both shoulders equally affected?	☐ Yes ☐ No, right better than left	☐ No, right worse than left ☐ Not applicable
v) Right Elbow	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
vi) Left Elbow	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
vii) Are both elbows equally affected?	☐ Yes ☐ No, right better than left	☐ No, right worse than left ☐ Not applicable
viii) Right Wrist	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
ix) Left wrist	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
x) Are both wrists equally affected?	☐ Yes ☐ No, right better than left	☐ No, right worse than left ☐ Not applicable
xi) Right hand	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
xii) Left hand	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
xiii) Are both hands equally affected?	☐ Yes ☐ No, right better than left	☐ No, right worse than left ☐ Not applicable
xiv) Is there thumb-in-palm deformity?	☐ Yes	☐ No
If yes, specify (check all that apply)	☐ Right	☐ Left
xv) Is there a right hand anomaly?	☐ Yes	☐ No
If yes, specify:	☐ Camptodactyly ☐ Overlapping fingers ☐ Syndactyly	☐ Complete or partial missing finger or fingernail ☐ Missing skin creases ☐ Other: _____

Version: May 3rd, 2022

Study ID:	Date completed (mm/dd/yyyy):

xvi) Is there a left hand anomaly?	☐ Yes	☐ No
If yes, specify:	☐ Camptodactyly ☐ Overlapping fingers ☐ Syndactyly	☐ Complete or partial missing finger or fingernail ☐ Missing skin creases ☐ Other: _____
xvii) Neck	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
xviii) Spine	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
xix) If spinal involvement, specify deformity:	_____	
xx) Right hip	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
xxi) Left hip	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
xxii) Are both hips equally affected?	☐ Yes ☐ No, right better than left	☐ No, right worse than left ☐ Not applicable
xxiii) Right knee	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
xxiv) Left knee	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
xxv) Are both knees equally affected?	☐ Yes ☐ No, right better than left	☐ No, right worse than left ☐ Not applicable
xxvi) Right foot	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
xxvii) Left foot	☐ No involvement ☐ Mild involvement	☐ Moderate involvement ☐ Severe involvement
xxviii) Are both feet equally affected?	☐ Yes ☐ No, right better than left	☐ No, right worse than left ☐ Not applicable
xxix) Is there a toe deformity on the right foot?	☐ Yes	☐ No
xxx) Is there a toe deformity on the left foot?	☐ Yes	☐ No

Clinical information. Co-morbid conditions:

Does your child have any co-morbid conditions (e.g. Intellectual disability, behavioral issues/diagnoses, learning difficulties, asthma, etc.)? ☐ **Yes** ☐ **No** ☐ **Don't know**

If yes, specify condition(s)- list all that apply:

_____

_____

_____

Clinical information. Prescribed medication:

Does your child take any prescribed medication on a daily basis? ☐ **Yes** ☐ **No** ☐ **Don't know**

If yes, list all prescription medications/substances taken (orally, inhaled, topically, IV, etc):

_____

_____

_____

Study ID:	Date completed (mm/dd/yyyy):

Clinical information. History of interventions specific to the upper extremity:

1. Was there shoulder surgery in the past? ☐ **Yes** ☐ **No** ☐ **Don't know**

If yes, indicate:

Surgery date (mm/dd/yyyy)	Shoulder surgical procedures (check all that apply per surgery date)	Side (check all that apply)
	☐ Bony procedures	☐ Right ☐ Left
________	☐ Soft tissue procedures	☐ Right ☐ Left
	☐ Capsular procedures	☐ Right ☐ Left
	☐ Other (describe):	☐ Right ☐ Left

Surgery date (mm/dd/yyyy)	Shoulder surgical procedures (check all that apply per surgery date)	Side (check all that apply)
	☐ Bony procedures	☐ Right ☐ Left
________	☐ Soft tissue procedures	☐ Right ☐ Left
	☐ Capsular procedures	☐ Right ☐ Left
	☐ Other (describe):	☐ Right ☐ Left

2. Was there elbow surgery in the past? ☐ **Yes** ☐ **No** ☐ **Don't know**

If yes, indicate:

Surgery date (mm/dd/yyyy)	Elbow surgical procedures (check all that apply per surgery date)	Side (check all that apply)
	☐ Bony procedures (e.g. humeral derotation)	☐ Right ☐ Left
________	☐ Soft tissue procedures (e.g. triceps tendon lengthening or transfer, pectoralis major transfer, latissiumus teres transfer, Steindler flexorplasty)	☐ Right ☐ Left
	☐ Capsular procedures (e.g. posterior release for flexion/arthrolysis)	☐ Right ☐ Left
	☐ Other (describe):	☐ Right ☐ Left

Surgery date (mm/dd/yyyy)	Elbow surgical procedures (check all that apply per surgery date)	Side (check all that apply)
	☐ Bony procedures (e.g. humeral derotation)	☐ Right ☐ Left
________	☐ Soft tissue procedures (e.g. triceps tendon lengthening or transfer, pectoralis major transfer, latissiumus teres transfer, Steindler flexorplasty)	☐ Right ☐ Left
	☐ Capsular procedures (e.g. posterior release for flexion/arthrolysis)	☐ Right ☐ Left
	☐ Other (describe):	☐ Right ☐ Left

Surgery date (mm/dd/yyyy)	Elbow surgical procedures (check all that apply per surgery date)	Side (check all that apply)
	☐ Bony procedures (e.g. humeral derotation)	☐ Right ☐ Left
	☐ Soft tissue procedures (e.g. triceps tendon lengthening or transfer, pectoralis major transfer, latissiumus teres transfer, Steindler flexorplasty)	☐ Right ☐ Left
________	☐ Capsular procedures (e.g. posterior release for flexion/arthrolysis)	☐ Right ☐ Left
	☐ Other (describe):	☐ Right ☐ Left

Study ID:	Date completed (mm/dd/yyyy):

Surgery date (mm/dd/yyyy)	Elbow surgical procedures (check all that apply per surgery date)	Side (check all that apply)
__________	☐ Bony procedures (e.g. humeral derotation)	☐ Right ☐ Left
	☐ Soft tissue procedures (e.g. triceps tendon lengthening or transfer, pectoralis major transfer, latissiumus teres transfer, Steindler flexorplasty)	☐ Right ☐ Left
	☐ Capsular procedures (e.g. posterior release for flexion/arthrolysis)	☐ Right ☐ Left
	☐ Other (describe):	☐ Right ☐ Left

3. Was there wrist surgery in the past? ☐ **Yes** ☐ **No** ☐ **Don't know**

If yes, indicate:

Surgery date (mm/dd/yyyy)	Wrist surgical procedures (check all that apply per surgery date)	Side (check all that apply)
__________	☐ Bony procedures (e.g. carpectomy, fusion)	☐ Right ☐ Left
	☐ Tendon transfers (e.g. FCU to ECU, FCU to ECRL, FCU to ECRB, ECU to ECRB, ECU to ECRL)	☐ Right ☐ Left
	☐ Soft tissue procedures (e.g. volar soft tissue release in forearm)	☐ Right ☐ Left
	☐ Capsular procedures (e.g. capsulotomy)	☐ Right ☐ Left
	☐ External fixator	☐ Right ☐ Left
	☐ Other (describe) (e.g. volar soft tissue release in foreram):	☐ Right ☐ Left

Surgery date (mm/dd/yyyy)	Wrist surgical procedures (check all that apply per surgery date)	Side (check all that apply)
__________	☐ Bony procedures (e.g. carpectomy, fusion)	☐ Right ☐ Left
	☐ Tendon transfers (e.g. FCU to ECU, FCU to ECRL, FCU to ECRB, ECU to ECRB, ECU to ECRL)	☐ Right ☐ Left
	☐ Soft tissue procedures (e.g. volar soft tissue release in forearm)	☐ Right ☐ Left
	☐ Capsular procedures (e.g. capsulotomy)	☐ Right ☐ Left
	☐ External fixator	☐ Right ☐ Left
	☐ Other (describe) (e.g. volar soft tissue release in foreram):	☐ Right ☐ Left

Surgery date (mm/dd/yyyy)	Wrist surgical procedures (check all that apply per surgery date)	Side (check all that apply)
__________	☐ Bony procedures (e.g. carpectomy, fusion)	☐ Right ☐ Left
	☐ Tendon transfers (e.g. FCU to ECU, FCU to ECRL, FCU to ECRB, ECU to ECRB, ECU to ECRL)	☐ Right ☐ Left
	☐ Soft tissue procedures (e.g. volar soft tissue release in forearm)	☐ Right ☐ Left
	☐ Capsular procedures (e.g. capsulotomy)	☐ Right ☐ Left
	☐ External fixator	☐ Right ☐ Left
	☐ Other (describe) (e.g. volar soft tissue release in foreram):	☐ Right ☐ Left

Surgery date (mm/dd/yyyy)	Wrist surgical procedures (check all that apply per surgery date)	Side (check all that apply)
__________	☐ Bony procedures (e.g. carpectomy, fusion)	☐ Right ☐ Left
	☐ Tendon transfers (e.g. FCU to ECU, FCU to ECRL, FCU to ECRB, ECU to ECRB, ECU to ECRL)	☐ Right ☐ Left
	☐ Soft tissue procedures (e.g. volar soft tissue release in forearm)	☐ Right ☐ Left
	☐ Capsular procedures (e.g. capsulotomy)	☐ Right ☐ Left
	☐ External fixator	☐ Right ☐ Left
	☐ Other (describe) (e.g. volar soft tissue release in foreram):	☐ Right ☐ Left

Study ID:	Date completed (mm/dd/yyyy):

4. Was there thumb surgery in the past? ☐ **Yes** ☐ **No** ☐ **Don't know**

If yes, indicate:

Surgery date (mm/yyyy)	Thumb surgical procedures (check all that apply per surgery date)	Side (check all that apply)
___________	☐ Bony procedures (e.g. metacarpophalangeal arthrodesis)	☐ Right ☐ Left
	☐ Soft tissue procedures (e.g. tendon release, first webspace release, flexor pollicis longus lengthening, advance ulnar collateral ligament)	☐ Right ☐ Left
	☐ Other (describe):	☐ Right ☐ Left

Surgery date (mm/yyyy)	Thumb surgical procedures (check all that apply per surgery date)	Side (check all that apply)
___________	☐ Bony procedures (e.g. metacarpophalangeal arthrodesis)	☐ Right ☐ Left
	☐ Soft tissue procedures (e.g. tendon release, first webspace release, flexor pollicis longus lengthening, advance ulnar collateral ligament)	☐ Right ☐ Left
	☐ Other (describe):	☐ Right ☐ Left

Surgery date (mm/yyyy)	Thumb surgical procedures (check all that apply per surgery date)	Side (check all that apply)
___________	☐ Bony procedures (e.g. metacarpophalangeal arthrodesis)	☐ Right ☐ Left
	☐ Soft tissue procedures (e.g. tendon release, first webspace release, flexor pollicis longus lengthening, advance ulnar collateral ligament)	☐ Right ☐ Left
	☐ Other (describe):	☐ Right ☐ Left

5. Was there finger surgery in the past? ☐ **Yes** ☐ **No** ☐ **Don't know**

If yes, indicate:

Surgery date (mm/dd/yyyy)	Finger surgical procedures (check all that apply per surgery date)	Side (check all that apply)
___________	☐ Bony procedures (e.g. metacarpophalangeal arthrodesis)	☐ Right ☐ Left
	☐ Soft tissue procedures (e.g. tendon release, webspace release, advance ulnar collateral ligament)	☐ Right ☐ Left
	☐ Other (describe):	☐ Right ☐ Left

Surgery date (mm/dd/yyyy)	Finger surgical procedures (check all that apply per surgery date)	Side (check all that apply)
___________	☐ Bony procedures (e.g. metacarpophalangeal arthrodesis)	☐ Right ☐ Left
	☐ Soft tissue procedures (e.g. tendon release, webspace release, advance ulnar collateral ligament)	☐ Right ☐ Left
	☐ Other (describe):	☐ Right ☐ Left

Surgery date (mm/dd/yyyy)	Finger surgical procedures (check all that apply per surgery date)	Side (check all that apply)
___________	☐ Bony procedures (e.g. metacarpophalangeal arthrodesis)	☐ Right ☐ Left
	☐ Soft tissue procedures (e.g. tendon release, webspace release, advance ulnar collateral ligament)	☐ Right ☐ Left
	☐ Other (describe):	☐ Right ☐ Left

Study ID:	Date completed (mm/dd/yyyy):

6.Were there surgical amputations in the past? ☐ **Yes** ☐ **No** ☐ **Don't know**

If yes, indicate:

Surgery date (mm/dd/yyyy)	Amputation (check all that apply per surgery date)	Side (check all that apply)
_____	☐ Upper extremity, indicate level: _____	☐ Right ☐ Left
	☐ Lower extremity, indicate level: _____	☐ Right ☐ Left

Clinical information. Does your child currently wear splints / braces? ☐ Yes ☐ No ☐ Don't know
If yes, check all that apply:

Upper extremity	☐ Dynamic elbow brace with hinge. Wearing Schedule:	☐ R ☐ L
	☐ Wrist cock-up Wearing Schedule:	☐ R ☐ L
	☐ Resting hand Wearing Schedule:	☐ R ☐ L
	☐ Thumb spica Wearing Schedule:	☐ R ☐ L
	☐ Other (specify splint): _____ Wearing Schedule:	☐ R ☐ L
Lower extremity	☐ AFO, articulated	☐ R ☐ L
	☐ AFO, non-articulated/solid	☐ R ☐ L
	☐ KAFO with knee lock	☐ R ☐ L
	☐ KAFO, no knee lock	☐ R ☐ L
	☐ HKAFO, lock	☐ R ☐ L
	☐ HKAFO, no lock	☐ R ☐ L
	☐ UCBL/UCB	☐ R ☐ L
	☐ Other (specify): _____	☐ R ☐ L
Spine	☐ TLSO (thoracic lumbar sacral orthosis)	
	☐ Nighttime bending braces	

Clinical information. What is the child's current main mode of mobility?
(check one answer that represents the child`s mobility best)

☐ Walk	☐ No aides
	☐ Lower extremity braces
	☐ Walker
	☐ Canes
	☐ Crutches
	☐ Other: _____
OR	
☐ Wheelchair	☐ Manual, self-propelled
	☐ Manual, pushed
	☐ Power/electric
OR	
☐ Crawl	

Study ID:	Date completed (mm/dd/yyyy):

OR	
☐ Stroller	

Clinical information. What is the child's current level of independence in self-care? (check one)

☐ Independent (i.e., able to complete self-care with no assistance)
☐ Independent with minimal assistance (i.e., able to complete self-care with minimal assistance from a person or device)
☐ Dependent (i.e., requires moderate to maximum assistance from a person or device)

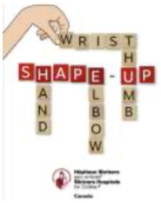
Rehabilitation (type, frequency, duration). Is your child receiving therapy for their upper limb (for example: occupational therapy, physical therapy, other)?

☐ No
☐ Yes, please describe:

- Specialty: ☐ occupational therapy ☐ physical therapy ☐ other:_____
- Frequency: number of times per week: _____/per week
- Duration (length of session in minutes): _____minutes/per session
- Type of therapeutic intervention. Please check all that apply:

  ☐ Dynamic/functional splinting (e.g exoskeletons, playskin)
  ☐ Assistive device/equipment (dressing tree, sock aid)
  ☐ Stretching program
  ☐ Muscle strengthening program
  ☐ other_____
  _____

Appendix 10. SHAPE-UP Administration Manual



Shriners Hospital Arthrogryposis Pediatric Evaluation – Upper Extremity
(SHAPE-UP)
Manual for Administration and Scoring

These instructions provide an overview of the new upper extremity (UE) measure for children with arthrogryposis multiplex congenita (AMC) called the SHAPE-UP and guidelines for its administration and scoring.

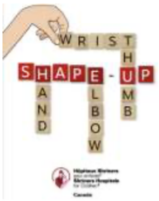
Table of Contents

I. Introduction

Background

A review of the literature revealed that generic upper extremity evaluation tools do not address the unique complexities associated with the contractures and the reduced active and passive range of motion associated with AMC. In the past, surgeons and therapists assessed a child's upper extremity function through clinical observation, anecdotal reports, range of motion and strength assessments. Surgical decision-making and measuring change over time were documented with narrative rather than evidence (Cachecho et al, 2022).

The need for a condition-specific measure to assess the upper extremity function of children with AMC was vocalized at the third International Arthrogryposis Symposium (Dahan-Oliel et al, 2019). Therapists, surgeons and families welcomed a measure that would assist in tracking upper extremity function, choosing interventions and documenting outcomes. The SHAPE-UP was developed based on the World Health Organization's International Classification of Functioning,

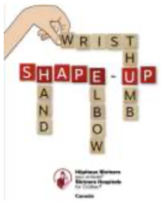


Health and Disability (ICF) framework. The SHAPE-UP describes the impairments, activity limitations, and participation restrictions in the performance of daily tasks in children with AMC.

Objectives
The SHAPE-UP was developed in 2022 to evaluate upper extremity function of children with AMC ages 1 - 21 years to ensure therapies are effective and evidence-based. The SHAPE-UP is the first standardized outcome measure specifically developed for children and youth with AMC. The SHAPE-UP focuses on task completion (i.e., if and how a particular task is completed) and the analysis of the motion and position of the joints involved in the completion of tasks (i.e., shoulder, elbow, forearm, wrist, thumb and fingers). The SHAPE-UP is video-based allowing it to be scored when viewed later. During administration of the test, the client can complete all tasks in a dynamic fashion without starting and stopping for the evaluator to record data.

Development
Approximately 40 experts from the AMC community were invited to participate in an electronic modified Delphi technique (MDT) via email to establish the different domains and associated items to be included in the item pool. The MDT is a process that begins with a set of carefully selected items. The final "item pool" was chosen from the scoping review of the literature on measures of upper extremity function, consultation with parents, and the results of the Delphi technique.

Twenty-three of the experts invited to participate completed the MDT. Clinicians participating in the Delphi were asked to complete separate rounds of the survey until 80% consensus was reached over a 6-month period. Experts were presented a series of items and asked to report on item clarity and item informativeness on a clearly defined 4-point Likert Scale. Additional comments were also sought regarding wording of items and item relevance.
An 80% agreement as a cut-off to accept or omit an item achieves content validity when at least 10 experts participate in the consensus development was used for retaining items for the next round. Responses of the first Delphi round informed two subsequent rounds at which time consensus was reached for all items. Based on the results of this Delphi process, a prototype of the SHAPE-UP was created with standardized instructions and materials.



II. General Instructions for Administration

Assigning a Participant ID Number:
The CRC recruiting the participant will assign a chronological study ID number as participants are recruited.

This study ID number is composed of the code of the establishment (e.g. CAN), followed by the unique number assigned to the child. The number of the first child registered in each establishment will be 01. The others will follow consecutively.

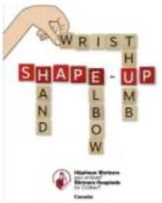
For example, the first participant at the Montreal site will be CAN01. The following participants will be assigned study ID numbers: CAN02, CAN03, CAN04…etc.

Establishment codes:
CAN= Canada
CHI= Chicago
GRE= Greenville
NCA= Northern California
PHL= Philadelphia
POR= Portland

Test environment
The SHAPE-UP is administered individually in a room that is free of distractions, large enough for throwing a ball and equipped with a small table. An 8' x 8' area is adequate.

Videotaping
The SHAPE-UP is recorded with any standard video recorder and a tripod. Sound recording (audio) is not needed. Set up the video recorder such that the child's upper extremity joints are clearly viewed. Be sure to locate the child and materials such that the movements can be clearly seen from the position of the videorecorder.

Preparation and Position of Child
Due to the nature of AMC and contractures possible, the child should be assessed in his/her most comfortable and typical position when completing a task. All items are placed on the table, presented at midline, without reference to which hand should be used unless otherwise noted. The suggested sequence of tasks does not have to be in the order of the tasks presented on the scoring form. Prior to starting the administration of the SHAPE-UP, explain the reason for the test. Example: "I am going to ask you to do some simple activities to better understand how you



do things with your arms and hands, like string beads, throw a ball. Show me how you usually/normally/typically would do it.

In order to ensure that the SHAPE-UP can be adequately scored after administration, please ensure that during recording of the session, there are no obstructions, such as the child's hair is up, remove long sleeved shirts, etc.

SHAPE-UP administration and scoring

Administration
The SHAPE-UP is administered by the site CRC/therapist. The Administration/Vignette case report form is used to collect:

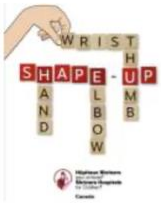
1. Basic demographic information: study ID, date of birth, date of test completion and total testing time.
2. Clinical information (questions 1-6) about the subject`s current use of upper extremities, preferred hand dominance and overall function.
3. Instructions for administration of the 11 tasks and a section for remarks and observations of subjects performance during each task (question 7) including description of pinch, finger/wrist deviation, use of technical and additional remarks/comments about ROM/joint position that may be difficult to view on the video or challenging to see.

The sequence on the form is the sequence recommended for administration.
Administer the task only once (as much as possible) to facilitate the scoring post administration.
Administration time is approximately 30 minutes depending on the child's age, level of ability, ease of following instructions and type of arthrogryposis

Scoring
The SHAPE-UP videos are scored centrally at the lead site by a group of therapists and study investigators who will review the videos from each site. This is to ensure consensus and agreement of scoring. The SHAPE-UP Scoring Case Report form is comprised of two sections:

Section I. Descriptive questions. This section consists of 5 questions regarding current function, personal goals and specific questions regarding the child's use of their upper extremities during mobility and transfers. This information is transferred from the Administration/Vignette Case Report Form completed and submitted by the participating sites.

Section II. Administration and Scoring of Tasks. This section describes the 11 tasks the child is asked to perform and the scoring method for the subtasks.



The 11 tasks of the SHAPE-UP are divided into subtasks (3-9 subtasks per task depending on the complexity of the task). The central evaluator observes how the child to complete the task in question and scores:

1- The child's ability to complete the task
2- the position of the key joints needed for each particular subtask or the movement used to complete the task

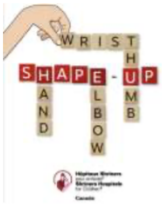
. Not every joint is analyzed in every subtask. The joint of interest for a particular subtask is indicated on the scoring form with a small icon/graphic.

The evaluator scores each subtask as follow:

- **Side:** circle R or L indicating the side the child primarily uses when completing the subtask, particularly for bilateral tasks such as opening jar, stringing beads, cutting paper.

- **Task completion score:** according to the Task Completion (see Table below.)
  - score NA if the child is too young to perform the subtask
  - The child scores a 3 (completion of task passively) when using external aid, assistance, compensatory movements, or a device. For example, if the child swings their arm and extends their trunk to reach their mouth, that would be a score of 3. Also, if a child were to use assistance from a table to passively push their arm up to reach their mouth that would also be a score of 3.

- **Position and motion of the joints:** required for each subtask according to the Analysis of Joint Motion and Position (see Table below) by circling one of the 3 choices provided in each box. Greyed sections do not require analysis.
  - When completing this section, it is important to reflect: "Does the child demonstrate the movement X required to complete the task". For example, when putting on a T-shirt, if the child goes through pronation and supination at some point in the task then they would get the highest/best score as they are displaying the highest-ranked position.
  - Some hierarchical scores are reflected as position and others as motion (shoulder internal rotation, neutral, external rotation versus no shoulder movement, partial shoulder movement, and full shoulder movement). This was implemented as some items require the child be in different positions in order to complete the task.
- **Compensatory movements:** the use of other body parts, using a particular starting position or adaptations such as larger zipper pull) that the child may use to complete the



task are described in the comment box on the Administration/Vignette Case Report Form.

Table 1.

Task Completion
NA= not applicable due to age of the child
0 = **unable** – The child is unable to complete any component of the task
1= **partial** completion of task **passively** – The child can partially complete the task using passive range of motion.
2= **partial** completion of task **actively** – The child can partially complete the task using active range of motion.
3= **completion** of task **passively** – The child can complete the task using passive range of motion. assistance of a body part, compensatory mechanisms, and/or an assistive device
4= **completion** of task **actively** – The child can complete the task using active range of motion.

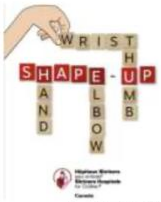
Table 2.

Analysis of Joint Motion and Position					
Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
Thumb in palm/ Thumb aligned with fingers/ Thumb open	No Finger Movement/ Partial Finger Movement/ Full Finger Movement	Extension/ Neutral/ Flexion	Supination/ Neutral/ Pronation	No Flexion/ Partial flexion/ Full flexion	No Shoulder Movement/ Partial Shoulder Movement/ Full Shoulder Movement

Description of the SHAPE-UP tasks

General Comments:

- Rate task completion based on whether or not the subtask is completed and whether with active or passive range. Do not score according to your knowledge of the child's range, rather what they are doing during the subtask
- The options for analysis of position or movement are presented hierarchically. If the best option (3rd) is observed even only briefly, rate the child with this best score.
- If the child uses swinging motion, arching of back or other body action to attempt/complete the task, score 3. These are considered compensatory movements.



- For the ball tasks, if child is using gravity to extend at the shoulder/throw ball, max task completion score is 2 (passive).
- Some study materials may be replaced by items that are lighter (plastic fork) or stiffer (pipe cleaner instead of string) to allow child to attempt/partially complete subtask. See Tips below.

1. **Pick up a cheerio, bring it to your mouth, place it back down in front of you, and let it go.**
   Tip: try and administer only 1 cheerio
   Focus: movement at the **Thumb/Finger/Elbow**
   2nd Goal: to describe the pinch/grasp pattern used for a small object by the child.
   ICF domain: activity

2. **Grasp a water bottle, bring it to your mouth, place it back down in front of you, and let it go.**

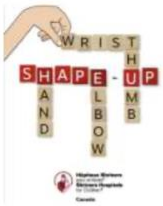
   Tip: water bottle may be emptied if child unable to complete due to weight
   Focus: movement at the **Thumb/Finger/Elbow/Shoulder**
   2nd Goal: to describe the grasp pattern used for medium sized object by the child.
   ICF domain: activity

3. **Open the jar, pour out a few beads, pick up bead and string 3 beads onto string, and close the jar.**

   Tip: String maybe replaced by pipe cleaner. Describe any adaptation used.
   Positional/movement analysis not required for subtask of stringing
   Focus: movement at the **Thumb/Finger/Wrist/Forearm**
   2nd Goal: to describe the movements used by the child during a bilateral task
   ICF domain: activity

4. **Pick up the marker, draw a circle or lines or write your name on this piece of paper with the marker, then cut or snip the paper (2-3 cuts in paper) using the scissors.**

   Tip:
   Focus: movement at the **Thumb/Finger/Wrist/Forearm**
   2nd Goal: to describe the movements used by the child during a task requiring 2 hands and used in school based environment
   ICF domain: activity, participation
   Detail: evaluator to remove cap from marker, Paper size 8.5x 11



5. **Pick up the fork, stick it into the Play-Doh, and bring fork to mouth.**

   Tip: plastic fork may replace metal fork
   Focus: movement at the **Thumb/Fingers/Wrist/Forearm/ Elbow/ Shoulder**
   2nd Goal: to describe the movements used by the child during a feeding task
   ICF domain: activity
   Detail: Evaluator places fork and container of Play-Doh in front of child.

6. **Put on and take off this t-shirt.**

   Tip: this is a bilateral task. Score right and left separately
   Focus: movement at the **Forearm/Elbow/Shoulder**
   2nd Goal: to describe the movements used by the child during a simple dressing task
   ICF domain: activity
   Detail: T-shirts (S-M-L are provided, choose item appropriate to child`s size.

7. **Put on this vest, attach the zipper, pull the zipper all the way up and then back down.**

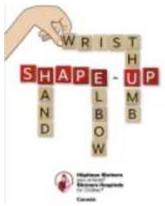
   Tip: this is a bilateral task. Score right and left separately
   Focus: movement at the **Thumb/Fingers/Forearm/Elbow/Shoulder**
   2nd Goal: to describe the movements used by the child during a complex dressing task
   ICF domain: activity
   Detail: vest with a zipper provided (S-M-L), choose item appropriate to child`s size.

8. **Put on this sock.**

   Tip: this is a bilateral task. Score right and left separately
   Focus: movement at the **Thumb/Fingers/Forearm//Elbow**
   2nd Goal: to describe the movements used by the child during a simple dressing task
   ICF domain: activity
   Detail: the child can remove and put on their own sock. If the child arrives in sandals, the evaluator will provide a loose fitting sock

9. **Throw the tennis ball underhand. Repeat task, throwing ball overhead.**

   Focus: movement at the **Wrist/Forearm/Elbow/Shoulder**
   2nd Goal: to describe the movements used by the child during play with small balls.
   ICF domain: participation
   Detail: includes wind up and throwing for underhand and overhand throw.



10. Throw the dodgeball underhand. Repeat task, throwing dodgeball overhead.

Tip: this is a Bilateral task. Score right and left separately
Focus: movement at the **Wrist/Forearm/Elbow/Shoulder**
2nd Goal: to describe the movements used by the child during play with medium sized balls.
ICF domain: participation
Detail: includes wind up and throwing for underhand and overhand throw.

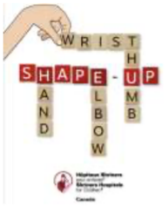
11. Pull down the shorts, reach your bottom from the back while holding 2 squares of toilet paper, also reach between your legs from the front while holding 2 squares of toilet paper, and pull the shorts back up.

Tip: removing shorts is a bilateral subtask. Score right and left separately
Focus: movement at the **Thumb/Fingers/Wrist/Forearm/Elbow/Shoulder**
2nd Goal: to describe the movements used by the child during a personal hygiene task
ICF domain: activity
Detail: use the paper shorts provided in the kit (S-M-L) appropriate to child's size.

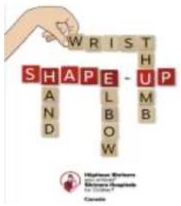
Testing Materials

- Cheerios
- 500 ml plastic Water

- Jar with 3 beads and string
- Chunky Marker 
- scissors
- Fork (*metal*) and playdough (individual containers provided)



- Tennis ball
- Medium ball (dodge ball size)
- T-shirt
- sock
- Vest with a zipper (S- M-_L)
- Paper shorts with elastic waist pants (*S-M-L*)
- Kleenex



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Appendix 11. SHAPE-UP Scoring Table (Original)

MEASURE FOR THE UPPER EXTREMITY IN ARTHROGRYPOSIS: DEVELOPMENT AND PSYCHOMETRICS – SHRINERS HOSPITALS ARTHROGRYPOSIS PEDIATRIC EVALUATION – UPPER EXTREMITY (SHAPE-UP)

SCORING SHEET

The purpose of the SHAPE-UP is to assess upper extremity (UE) function in children with AMC with UE involvement. The SHAPE-UP will describe the impairments, activity limitations, and participation restrictions in the performance of daily tasks in order to guide treatment decision-making and evaluate treatment effectiveness.

Section I. Descriptive questions.

1. What upper extremity movements are difficult for you/your child to do and/or prevent the completion of an everyday task? (*Please indicate who reports.)

☐ Child ☐ Parent

2. What would you like to do that you cannot do now?

 Child: _____

 Parent: _____

3. Please describe any technical aids or modifications that your child requires to complete daily tasks involving the upper limbs (ex plastic utensils, button hook, sock aid.

_____

4. Does your child use their arms to / Do you use your arms to :

- Propel a manual wheelchair ☐ Yes ☐ No. If yes, describe _____

_____

- Operate a motorized wheelchair ☐ Yes ☐ No. If yes, describe _____

_____

- Use a walking aid (cane, crutch, walker) ☐ Yes ☐ No. If yes, describe _____

_____

-Other: _____ ☐ Yes ☐ No. If yes, describe _____

5. Does the child use their arms to perform the following transfers:

Transfers	Uses their arms		Cannot use their arms	Cannot complete due to LE contracture	Describe
	Right	Left			
Bed positioning					
Lying to sitting					
Sitting to standing / Standing to sitting					
Toilet transfer					
Bathtub/shower transfer					
Getting in and out of a car					
Other:					
Other:					
Other:					

Section II. Administration and Scoring of Tasks

Scoring:	
Task Completion –Circle the number which best describes the method of completion	Analysis of Joint Movement and /or Position
NA= not applicable if the child did not perform the task	
0 = The child is **unable** to attempt to complete or complete any part of the subtasks.	The position and movement of the upper extremity joints involved in the task are described and rated from the least to most optimal position/movement during completion of the subtasks.
1= The child scores a 1 when they are **attempting** to complete the task with **minimal success**.	
2= The child scores a 2 when they can **actively partially** complete the task with any **physical assistance**.	
3= The child scores a 3 when they can **passively complete** the task when using external aid (not physical aid from a person), compensatory movements, or a device	
4= The child scores a 4 when they can **actively complete** the task without any external aid or compensatory movement.	

Please indicate child's dominance/preferred hand: ☐ Right ☐ Left ☐ Neither/no preference

General Instructions

- Prior to starting, explain what you will be asking the child to do. Example: "I am going to ask you to do some simple activities, like string beads, things like that. Show me how you usually/normally/typically would do it.
- Age: please use your clinical judgement as to whether the child is developmentally ready/able to complete the task. For example, children are generally able to string beads at 2 years of age. If the child is too young to perform the task, please score NA.
- R/L: please indicate which side the child uses for each subtask,
- For bilateral tasks (6, 7, 8, 10, 11) – donning T-shirt, jacket, sock, throwing basketball, removing shorts, certain subtasks are usually completed with both hands. Please score these subtasks for right and left sides as indicated with separate rows for right and left.
- For grasping tasks (item 1- cheerio, 2-water bottle, 3- bead) complete both right and left side only when child uses both hands for these tasks in order to differentiate between those bilateral tasks requiring asymmetrical movement such as the zipper. Describe in comment box type of grasp used - example: scissor grasp vs hands side-to-side vs hands with fingers facing.
- For the first 3 tasks please indicate if ulnar or radial deviation is present in the wrist or fingers.

1. Pick up a cheerio, bring it to your mouth, place it back down in front of you, and let it go.
   *Select R **OR** L in column "Arm" to describe the side the child primarily uses throughout each subtask. If both hands are used to grasp, complete R **AND** L rows.*

Arm	Item	Task completion						Analysis of Joint Motion and Position					
								Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
■ R ☐ L	Pick up cheerio	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Fingers flexed (hand fisted) ☐ Hand opened (not fisted) ☐ Grasp between thumb and fingers (opposition)	☐ Extension ☐ Flexion ☐ Neutral			
☐ R ■ L	Pick up cheerio	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Fingers flexed (hand fisted) ☐ Hand opened (not fisted) ☐ Grasp between thumb and fingers (opposition)	☐ Extension ☐ Flexion ☐ Neutral			
☐ R ☐ L	Bring it to mouth	NA	0	1	2	3	4				☐ Pronation ☐ Neutral ☐ Supination	☐ No flexion ☐ Partial flexion ☐ Full flexion	☐ No shoulder movement ☐ Partial shoulder movement ☐ Full shoulder movement
☐ R ☐ L	Place it back down in front of you	NA	0	1	2	3	4				☐ Supination ☐ Neutral ☐ Pronation	☐ No extension ☐ Partial extension ☐ Full extension	
☐ R ☐ L	Let it go	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Fingers flexed (hand fisted) ☐ Hand opened (not fisted) ☐ Grasp between thumb and fingers (opposition)	☐ Extension ☐ Flexion ☐ Neutral			

Remarks: please describe use of opposite arm, and any compensatory movements (such as a particular starting position or assistive device such as sock aid, larger zipper pull, etc.) that the child may use to complete the task.
Describe pinch/grasp pattern
Describe any deviation (ulnar or radial) if present.

2. Grasp a water bottle, bring it to your mouth, place it back down in front of you, and let it go.
*Select R **OR** L in column "Arm" to describe the side the child primarily uses throughout each subtask. If both hands are used to grasp, complete R **AND** L rows.*

Arm	Item	Score						Analysis of Joint Motion and Position					
								Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
■ R ☐ L	Grasp water bottle	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Fingers flexed (hand fisted) ☐ Hand opened (not fisted) ☐ Grasp between thumb and fingers (opposition)	☐ Flexion ☐ Neutral ☐ Extension	☐ Supination ☐ Pronation ☐ Neutral		
☐ R ■ L	Grasp water bottle	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Fingers flexed (hand fisted) ☐ Hand opened (not fisted) ☐ Grasp between thumb and fingers (opposition)	☐ Flexion ☐ Neutral ☐ Extension	☐ Supination ☐ Pronation ☐ Neutral		
☐ R ☐ L	Bring it to mouth	NA	0	1	2	3	4			☐ Flexion ☐ Neutral ☐ Extension	☐ Supination ☐ Pronation ☐ Neutral	☐ No flexion ☐ Partial flexion ☐ Full flexion	☐ No shoulder movement ☐ Partial shoulder movement ☐ Full shoulder movement
☐ R ☐ L	Place it back down in front of you	NA	0	1	2	3	4			☐ Flexion ☐ Neutral ☐ Extension	☐ Supination ☐ Pronation ☐ Neutral	☐ No extension ☐ Partial extension ☐ Full extension	
☐ R ☐ L	Let it go	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Fingers flexed (hand fisted) ☐ Hand opened (not fisted) ☐ Grasp between thumb and fingers (opposition)	☐ Flexion ☐ Neutral ☐ Extension	☐ Supination ☐ Pronation ☐ Neutral		

Remarks: please describe any compensatory movements: use opposite arm, a particular starting position (standing versus sitting or assistive device such as sock aid, larger zipper pull, etc.) that the child may use to complete the task.
Describe pinch/grasp pattern
Describe any deviation (ulnar or radial) if present.

3. Open the jar, pour out a few beads, pick up bead and string 3 beads onto string, and close the jar. *Select R **OR** L in column "Arm" to describe the side the child primarily uses throughout each subtask. If both hands are used to grasp, complete R **AND** L rows.*

ARM	Item	Score						Analysis of Joint Motion and Position					
								Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
☐ R ☐ L	Stabilizes jar to open	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ No Finger Movement ☐ Partial Finger Movement ☐ Full Finger Movement	☐ Extension ☐ Flexion ☐ Neutral	☐ Supination ☐ Pronation ☐ Neutral		
☐ R ☐ L	Open jar	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ No Finger Movement ☐ Partial Finger Movement ☐ Full Finger Movement	☐ Extension ☐ Flexion ☐ Neutral	☐ Supination ☐ Neutral ☐ Pronation		
☐ R ☐ L	Pour out	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ No Finger Movement ☐ Partial Finger Movement ☐ Full Finger Movement		☐ Supination ☐ Neutral ☐ Pronation		
☐ R ☐ L	Hold string	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ No Finger Movement ☐ Partial Finger Movement ☐ Full Finger Movement				
☐ R ☐ L	Pick up bead	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ No Finger Movement ☐ Partial Finger Movement ☐ Full Finger Movement				
■ R ■ L	String 3 beads	NA	0	1	2	3	4						
☐ R ☐ L	Stabilizes jar to close	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ No Finger Movement ☐ Partial Finger Movement ☐ Full Finger Movement	☐ Extension ☐ Flexion ☐ Neutral	☐ Supination ☐ Pronation ☐ Neutral		
☐ R ☐ L	Close jar	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ No Finger Movement ☐ Partial Finger Movement ☐ Full Finger Movement	☐ Extension ☐ Flexion ☐ Neutral	☐ Supination ☐ Neutral ☐ Pronation		

Remarks: please describe any compensatory movements: use of opposite arm, a particular starting position (standing versus sitting) or assistive device such as sock aid, larger zipper pull, etc.) that the child may use to complete the task.
Describe any deviation (ulnar or radial) if present. Describe pinch/grasp pattern.

4. Pick up the marker, draw a circle or lines or write your name on this piece of paper with the marker, then cut or snip the paper (2-3 cuts in paper) using the scissors.
Select R or L in column "Arm" to describe the side the child primarily uses throughout each subtask.
(Evaluator to place uncapped marker across the midline). Paper size 8.5x 11

ARM	Item		Score					Analysis of Joint Motion and Position					
								Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
☐ R ☐ L	Pick up the marker	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ No Finger Movement ☐ Partial Finger Movement ☐ Full Finger Movement	☐ Extension ☐ Flexion ☐ Neutral	☐ Supination ☐ Neutral ☐ Pronation		
☐ R ☐ L	Write name	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ No Finger Movement ☐ Partial Finger Movement ☐ Full Finger Movement	☐ Extension ☐ Flexion ☐ Neutral	☐ Supination ☐ Neutral ☐ Pronation		
☐ R ☐ L	Stabilize paper to cut	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ No Finger Movement ☐ Partial Finger Movement ☐ Full Finger Movement	☐ Extension ☐ Flexion ☐ Neutral	☐ Supination ☐ Pronation ☐ Neutral		
☐ R ☐ L	Cut paper	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ No Finger Movement ☐ Partial Finger Movement ☐ Full Finger Movement	☐ Flexion ☐ Extension ☐ Neutral	☐ Supination ☐ Pronation ☐ Neutral		

Remarks: please describe any compensatory movements: use of opposite arm, a particular starting position (standing versus sitting) or assistive device such as sock aid, larger zipper pull, etc.)

5. Pick up the fork, stick it into the Play-Doh, and bring fork to mouth.
Select R or L in column "Arm" to describe the side the child primarily uses throughout each subtask.
(Evaluator places fork and 3 inch by 3-inch piece of Play-Doh in front of child).

Arm	Item	Score						Analysis of Joint Motion and Position					
								Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
☐ R ☐ L	Pick up the fork	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Hand opened (not fisted) ☐ Fingers flexed (hand fisted) ☐ Grasp between thumb and fingers (opposition)	☐ Extension ☐ Flexion ☐ Neutral	☐ Supination ☐ Neutral ☐ Pronation		
☐ R ☐ L	Stick fork into Playdoh	NA	0	1	2	3	4			☐ Extension ☐ Flexion ☐ Neutral		☐ No elbow movement ☐ Partial elbow movement ☐ Full elbow movement	
☐ R ☐ L	Bring fork to Mouth	NA	0	1	2	3	4				☐ Pronation ☐ Neutral ☐ Supination	☐ No elbow flexion ☐ Partial elbow flexion ☐ Full elbow flexion	

Remarks: please describe any compensatory movements: use of the other limb or opposite arm, a particular starting position (standing versus sitting) or assistive device such as sock aid, larger zipper pull, etc).

6. Put on and take off a t-shirt.
Starting position: Therapist/child places t-shirt, scoring starts when child pushes arms through sleeves.
Note that the following items do not need to be completed in the presented order.

Arm	Item	Score						Analysis of Joint Motion and Position					
								Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
■ R ☐ L	Push arms through sleeves (extending)	NA	0	1	2	3	4				☐ Supination ☐ Neutral ☐ Pronation	☐ No elbow extension ☐ Partial elbow extension ☐ Full elbow extension	☐ No Shoulder Movement ☐ Partial Shoulder Movement ☐ Full Shoulder Movement
☐ R ■ L	Push arms through sleeves (extending)	NA	0	1	2	3	4				☐ Supination ☐ Neutral ☐ Pronation	☐ No elbow extension ☐ Partial elbow extension ☐ Full elbow extension	☐ No Shoulder Movement ☐ Partial Shoulder Movement ☐ Full Shoulder Movement
■ R ☐ L	Remove arms from sleeves (elbows flexed)	NA	0	1	2	3	4					☐ No elbow movement ☐ Partial elbow movement ☐ Full elbow movement	☐ No Shoulder Movement ☐ Partial Shoulder Movement ☐ Full Shoulder Movement
☐ R ■ L	Remove arms from sleeves (elbows flexed)	NA	0	1	2	3	4					☐ No elbow movement ☐ Partial elbow movement ☐ Full elbow movement	☐ No Shoulder Movement ☐ Partial Shoulder Movement ☐ Full Shoulder Movement

Remarks: please describe any compensatory movements: use of the other limb or opposite arm, a particular starting position (standing versus sitting) or assistive device such as sock aid, larger zipper pull, etc.)

7. Put on this vest, attach the zipper, and pull the zipper all the way up and then all the way back down.
   * If the child cannot put on or take off the vest, therapist can assist as needed
   *if child cannot reach zipper to pull down, therapist to position the zipper where child can attempt to pull down

Arm	Item	Score						Analysis of Joint Motion and Position					
								Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
■ R ☐ L	Put on item of clothing	NA	0	1	2	3	4						
☐ R ■ L	Put on item of clothing	NA	0	1	2	3	4						
☐ R ☐ L	Stabilize zipper	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Hand opened (not fisted) ☐ Fingers flexed (hand fisted) ☐ Grasp between thumb and fingers (opposition)		☐ Supination ☐ Neutral ☐ Pronation	☐ No elbow extension ☐ Partial elbow extension ☐ Full elbow extension	☐ External rotation ☐ Internal rotation ☐ Neutral
☐ R ☐ L	Attach zipper	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Hand opened (not fisted) ☐ Fingers flexed (hand fisted) ☐ Grasp between thumb and fingers (opposition)		☐ Supination ☐ Neutral ☐ Pronation	☐ No elbow extension ☐ Partial elbow extension ☐ Full elbow extension	☐ External rotation ☐ Internal rotation ☐ Neutral
☐ R ☐ L	Pull zipper 0-1/3 of the way up	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Hand opened (not fisted) ☐ Fingers flexed (hand fisted) ☐ Grasp between thumb and fingers (opposition)		☐ Supination ☐ Neutral ☐ Pronation	☐ No elbow flexion ☐ Partial elbow flexion ☐ Full elbow flexion	☐ Internal rotation ☐ External rotation ☐ Neutral
☐ R ☐ L	Pull zipper 0-2/3 of the way up	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Hand opened (not fisted) ☐ Fingers flexed (hand fisted) ☐ Grasp between thumb and fingers (opposition)		☐ Supination ☐ Neutral ☐ Pronation	☐ No elbow flexion ☐ Partial elbow flexion ☐ Full elbow flexion	☐ Internal rotation ☐ External rotation ☐ Neutral
☐ R ☐ L	Pull zipper all the way up	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Hand opened (not fisted) ☐ Fingers flexed (hand fisted) ☐ Grasp between thumb and fingers (opposition)		☐ Pronation ☐ Supination ☐ Neutral	☐ No elbow flexion ☐ Partial elbow flexion ☐ Full elbow flexion	☐ Internal rotation ☐ External rotation ☐ Neutral
☐ R ☐ L	Pull zipper 0-1/3 of the way down*	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Hand opened (not fisted) ☐ Fingers flexed (hand fisted) ☐ Grasp between thumb and fingers (opposition)		☐ Pronation ☐ Supination ☐ Neutral	☐ No elbow extension ☐ Partial elbow extension ☐ Full elbow Extension	☐ Internal rotation ☐ External rotation ☐ Neutral
☐ R ☐ L	Pull zipper 0-2/3 of the way down*	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Hand opened (not fisted) ☐ Fingers flexed (hand fisted) ☐ Grasp between thumb and fingers (opposition)		☐ Supination ☐ Neutral ☐ Pronation	☐ No elbow extension ☐ Partial elbow extension ☐ Full elbow Extension	☐ Internal rotation ☐ External rotation ☐ Neutral
☐ R ☐ L	Pull zipper all the way down*	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Hand opened (not fisted) ☐ Fingers flexed (hand fisted) ☐ Grasp between thumb and fingers (opposition)		☐ Supination ☐ Neutral ☐ Pronation	☐ No elbow extension ☐ Partial elbow extension ☐ Full elbow Extension	☐ Internal rotation ☐ External rotation ☐ Neutral

Remarks: please describe any compensatory movements: use of the other limb or opposite arm, a particular starting position (standing versus sitting) or assistive device such as sock aid, larger zipper pull, etc.).

8. Put on this sock.

Does a lower extremity contracture prohibit completion of the task YES ☐ NO ☐
Are the upper extremities used to complete task YES ☐ NO ☐

Arm	Item		Score					Analysis of Joint Motion and Position					
								Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
■ R ☐ L	Put on sock over toes	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Fingers flexed (hand fisted) ☐ Hand opened (not fisted) ☐ Grasp between thumb and fingers (opposition)		☐ Supination ☐ Neutral ☐ Pronation	☐ No elbow extension ☐ Partial elbow extension ☐ Full elbow Extension	
☐ R ■ L	Put on sock over toes	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Fingers flexed (hand fisted) ☐ Hand opened (not fisted) ☐ Grasp between thumb and fingers (opposition)		☐ Supination ☐ Neutral ☐ Pronation	☐ No elbow extension ☐ Partial elbow extension ☐ Full elbow Extension	
■ R ☐ L	Pull sock over heel	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Fingers flexed (hand fisted) ☐ Hand opened (not fisted) ☐ Grasp between thumb and fingers (opposition)		☐ Supination ☐ Neutral ☐ Pronation		
☐ R ■ L	Pull sock over heel	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Fingers flexed (hand fisted) ☐ Hand opened (not fisted) ☐ Grasp between thumb and fingers (opposition)		☐ Supination ☐ Neutral ☐ Pronation		

Remarks: please describe any compensatory movements: use of the other limb or opposite arm, a particular starting position (standing versus sitting) or assistive device such as sock aid, larger zipper pull, etc).

9. Throw the tennis ball underhand. Repeat task, throwing ball overhead.
Select R or L in column "Arm" to describe the side the child primarily uses throughout each subtask.
Evaluator hands ball to child.

Arm	Item	Score						Analysis of Joint Motion and Position					
								Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
☐ R ☐ L	Wind up in preparation to throw ball underhand (into extension position to throw)	NA	0	1	2	3	4			☐ Flexion ☐ Neutral ☐ Extension	☐ Pronation ☐ Neutral ☐ Supination	☐ No elbow extension ☐ Partial elbow extension ☐ Full elbow extension	☐ Flexion ☐ Neutral ☐ Extension
☐ R ☐ L	Throw ball underhand	NA	0	1	2	3	4			☐ Extension ☐ Neutral ☐ Flexion	☐ Pronation ☐ Neutral ☐ Supination	☐ No elbow extension ☐ Partial elbow extension ☐ Full elbow extension	☐ Extension ☐ Neutral ☐ Flexion
☐ R ☐ L	Wind up in preparation to throw ball overhead (into flexion position at the shoulder to throw)	NA	0	1	2	3	4			☐ Flexion ☐ Neutral ☐ Extension	☐ Supination ☐ Neutral ☐ Pronation	☐ No elbow flexion ☐ Partial elbow flexion ☐ Full elbow flexion	☐ Extension ☐ Neutral ☐ Flexion
☐ R ☐ L	Throw ball overhead (into extension posture at the shoulder when you release)	NA	0	1	2	3	4			☐ Extension ☐ Neutral ☐ Flexion	☐ Supination ☐ Neutral ☐ Pronation	☐ No elbow extension ☐ Partial elbow extension ☐ Full elbow extension	☐ Flexion ☐ Neutral ☐ Extension

Remarks: please describe any compensatory movements: use of opposite arm, a particular starting position (standing versus sitting) or assistive device such as sock aid, larger zipper pull, etc)

10. Throw the dodgeball overhead with both hands.
Evaluator hands ball to child

Arm	Item	Score						Analysis of Joint Motion and Position					
								Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
■ R ☐ L	Wind up in preparation to throw ball overhead (into flexion position at the shoulder to throw)	NA	0	1	2	3	4			☐ Flexion ☐ Neutral ☐ Extension	☐ Pronation ☐ Supination ☐ Neutral	☐ No elbow flexion ☐ Partial elbow flexion ☐ Full elbow flexion	☐ Extension ☐ Neutral ☐ Flexion
☐ R ■ L	Wind up in preparation to throw ball overhead (into flexion position at the shoulder to throw)	NA	0	1	2	3	4			☐ Flexion ☐ Neutral ☐ Extension	☐ Pronation ☐ Supination ☐ Neutral	☐ No elbow flexion ☐ Partial elbow flexion ☐ Full elbow flexion	☐ Extension ☐ Neutral ☐ Flexion
■ R ☐ L	Throw ball overhead (into extension posture at the shoulder when you release)	NA	0	1	2	3	4			☐ Extension ☐ Neutral ☐ Flexion	☐ Supination ☐ Pronation ☐ Neutral	☐ No elbow extension ☐ Partial elbow extension ☐ Full elbow extension	☐ Flexion ☐ Neutral ☐ Extension
☐ R ■ L	Throw ball overhead (into extension posture at the shoulder when you release)	NA	0	1	2	3	4			☐ Extension ☐ Neutral ☐ Flexion	☐ Supination ☐ Pronation ☐ Neutral	☐ No elbow extension ☐ Partial elbow extension ☐ Full elbow extension	☐ Flexion ☐ Neutral ☐ Extension

Remarks: please describe any compensatory movements: use of the opposite arm, a particular starting position (standing versus sitting) or assistive device such as sock aid, larger zipper pull, etc.).

11. Pull down the shorts, reach your bottom from the back while holding 2 squares of toilet paper, also reach between your legs from the front while holding 2 squares of toilet paper, and pull the shorts back up.

Arm	Item	Score						Analysis of Joint Motion and Position					
								Thumb	Fingers	Wrist	Forearm	Elbow	Shoulder
■ R ☐ L	Pull down shorts	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Fingers flexed (hand fisted) ☐ Hand opened (not fisted)/ ☐ Grasp between thumb and fingers (opposition)	☐ Flexion ☐ Extension ☐ Neutral		☐ No Elbow extension ☐ Partial Elbow extension ☐ Full Elbow extension	
☐ R ■ L	Pull down shorts	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Fingers flexed (hand fisted) ☐ Hand opened (not fisted)/ ☐ Grasp between thumb and fingers (opposition)	☐ Flexion ☐ Extension ☐ Neutral		☐ No Elbow extension ☐ Partial Elbow extension ☐ Full Elbow extension	
☐ R ☐ L	Reach buttocks area [back]	NA	0	1	2	3	4			☐ Extension ☐ Neutral ☐ Flexion	☐ Pronation ☐ Neutral ☐ Supination	☐ No Elbow extension ☐ Partial Elbow extension ☐ Full Elbow extension	☐ No Shoulder Movement ☐ Partial Shoulder Movement ☐ Full Shoulder Movement
☐ R ☐ L	Reach between legs [front]	NA	0	1	2	3	4			☐ Extension ☐ Neutral ☐ Flexion	☐ Supination ☐ Neutral ☐ Pronation	☐ No Elbow extension ☐ Partial Elbow extension ☐ Full Elbow extension	☐ No Shoulder Movement ☐ Partial Shoulder Movement ☐ Full Shoulder Movement
■ R ☐ L	Pull up shorts	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Fingers flexed (hand fisted) ☐ Hand opened (not fisted)/ ☐ Grasp between thumb and fingers (opposition)			☐ No Elbow flexion ☐ Partial Elbow flexion ☐ Full Elbow flexion	
☐ R ■ L	Pull up shorts	NA	0	1	2	3	4	☐ Thumb in palm ☐ Thumb aligned with fingers ☐ Thumb open	☐ Fingers flexed (hand fisted) ☐ Hand opened (not fisted)/ ☐ Grasp between thumb and fingers (opposition)			☐ No Elbow flexion ☐ Partial Elbow flexion ☐ Full Elbow flexion	

Remarks: please describe any compensatory movements: use of opposite arm, a particular starting position (standing versus sitting) or assistive device such as sock aid, larger zipper pull, etc.).

Appendix 12. SHAPE-UP Task Completion 5-Point Scale (Original)

Task Completion: Circle the number which best describes the method of completion	
NA= not applicable if the child did not perform the task	
0	The child is unable to attempt to complete or complete any part of the subtasks.
1	The child scores a 1 when they are attempting to complete the task with minimal success.
2	The child scores a 2 when they can actively partially complete the task with any physical assistance.
3	The child scores a 3 when they can passively complete the task when using external aid (not physical aid from a person), compensatory movements, or a device
4	The child scores a 4 when they can actively complete the task without any external aid or compensatory movement.
Higher scores denote better performance.	

Appendix 13. SHAPE-UP Scoring Completion 3-PointScale (Post-Rasch Analysis)

Task Completion Score	
0	Unable to complete any part of the task even when given assistance from caregiver
1	Able to complete task with compensatory movements, external aids, adaptive equipment or assistance from caregivers
2	Able to complete without external aid or compensatory movements

Appendix 14. SHAPE-UP Scoring Table (Post Rasch Analysis)

MEASURE FOR THE UPPER EXTREMITY IN ARTHROGRYPOSIS: DEVELOPMENT AND PSYCHOMETRICS – SHRINERS HOSPITALS ARTHROGRYPOSIS PEDIATRIC EVALUATION – UPPER EXTREMITY (SHAPE-UP)

SCORING SHEET

The purpose of the SHAPE-UP is to assess upper extremity (UE) function in children with AMC with UE involvement. The SHAPE-UP will describe the impairments, activity limitations, and participation restrictions in the performance of daily tasks in order to guide treatment decision-making and evaluate treatment effectiveness.

Section I. Descriptive questions.

6. What upper extremity movements are difficult for you/your child to do and/or prevent the completion of an everyday task? (*Please indicate who reports.)

☐ Child ☐ Parent

7. What would you like to do that you cannot do now?

 Child: _____

 Parent: _____

8. Please describe any technical aids or modifications that your child requires to complete daily tasks involving the upper limbs (ex plastic utensils, button hook, sock aid.

_____

9. Please indicate your child's dominance/preferred hand: ☐ Right ☐ Left ☐ Neither/no preference

10. Does your child use their arms to / Do you use your arms to :

- Propel a manual wheelchair ☐ Yes ☐ No. If yes, describe _____

_____

- Operate a motorized wheelchair ☐ Yes ☐ No. If yes, describe _____

_____

- Use a walking aid (cane, crutch, walker) ☐ Yes ☐ No. If yes, describe _____

_____

-Other: _____ ☐ Yes ☐ No. If yes, describe _____

11. Does the child use their arms to perform the following transfers:

Transfers	Uses their arms		Cannot use their arms	Cannot complete due to LE contracture	Describe
	Right	Left			
Bed positioning					
Lying to sitting					
Sitting to standing / Standing to sitting					
Toilet transfer					
Bathtub/shower transfer					
Getting in and out of a car					
Other:					
Other:					
Other:					

Section II. Administration and Scoring of Tasks

Scoring:	
Task Completion –Circle the number which best describes the method of completion	**Analysis of Joint Movement and /or Position**
NA= not applicable if the child did not perform the task	
0 = The child is **unable** to complete any part of the task even when given assistance from a caregiver.	The position and movement of the upper extremity joints involved in the task are described and rated.
1= The child scores a 1 when they are able to complete the task **with compensatory movements, external aids, adaptive equipment or assistance from a caregiver**.	
2= The child scores a 2 when they are able to complete the task **without external aid or compensatory movement**.	

General Instructions

- Prior to starting, explain what you will be asking the child to do. Example: "I am going to ask you to do some simple activities, like string beads, things like that. Show me how you usually/normally/typically would do it.
- Age: please use your clinical judgement as to whether the child is developmentally ready/able to complete the task. For example, children are generally able to string beads at 2 years of age. If the child is too young to perform the task, please score NA.
- R/L: please indicate which side the child uses for each subtask when indicated
- For bilateral tasks – beading, vest, throwing dodgeball, removing shorts, certain subtasks are usually completed with both hands. Please score these subtasks for right and left sides as indicated with separate rows for right and left.
- For grasping tasks complete both right and left side only when child uses both hands for these tasks in order to differentiate between those bilateral tasks requiring asymmetrical movement such as the zipper. Describe in comment box type of grasp used - example: scissor grasp vs hands side-to-side vs hands with fingers facing.

12. Pick up a cheerio, bring it to your mouth, place it back down in front of you, and let it go.

Arm	Subtask	Task Completion				Subtask Analysis of Joint Motion and Position						
						Prehension	Limitation				Adaptive Equipment/ Physical Assistance	Compensatory Movements
							Wrist	Forearm	Elbow	Shoulder		
☐ R ☐ L	Pick up cheerio	NA	0	1	2	☐ Scissor between the fingers ☐ Back of thumb and finger (fingers bending to thumb) ☐ Thumb in palm ☐ Pinch	☐ Stuck in flexion, minimal movement ☐ Flexed, some movement ☐ Stuck in extension, minimal movement ☐ Extended, some movement ☐ No limitation	☐ Too much pronation ☐ Too much supination ☐ No limitation	☐ Stuck in extension ☐ Some flexion/extension ☐ Stuck in flexion ☐ No limitation	☐ Too much internal rotation ☐ Some internal rotation ☐ No limitation	☐ Yes, describe ☐ No limitation	☐ Bimanual: ☐ Back of palm ☐ Palm to palm ☐ Crossover palm to palm ☐ Hand to forearm/chest ☐ Trunk ☐ Other body part:
☐ R ☐ L	Bring it to mouth	NA	0	1	2	☐ Scissor between the fingers ☐ Back of thumb and finger (fingers bending to thumb) ☐ Thumb in palm ☐ Pinch	☐ Stuck in flexion, minimal movement ☐ Flexed, some movement ☐ Stuck in extension, minimal movement ☐ Extended, some movement ☐ No limitation	☐ Too much pronation ☐ Too much supination ☐ No limitation	☐ Stuck in extension ☐ Some flexion/extension ☐ Stuck in flexion ☐ No limitation	☐ Too much internal rotation ☐ Some internal rotation ☐ No limitation	☐ Yes, describe ☐ No limitation	☐ Neck flexion ☐ Wrist flexion to reach ☐ Supination ☐ Passive elbow flexion ☐ Other body part:

Remarks: please describe any compensatory movements: use opposite arm, a particular starting position (standing versus sitting or assistive device such as sock aid, larger zipper pull, etc.) that the child may use to complete the task.

Describe pinch/grasp pattern (symmetrical or asymmetrical):

Describe any deviation (ulnar or radial) if present:

Describe any congenital anomaly (camptodactyly, pterygium)

13. Grasp a water bottle, bring it to your mouth, place it back down in front of you, and let it go.

Arm	Subtask	Task Completion				Subtask Analysis of Joint Motion and Position						
						Prehension	Limitation				Adaptive Equipment/ Physical Assistance	Compensatory Movements
							Wrist	Forearm	Elbow	Shoulder		
■ R □ L	Grasp water bottle	NA	0	1	2	□ Scissor between the fingers □ Back of thumb and finger (fingers bending to thumb) □ Thumb in palm □ Pinch	□ Stuck in flexion, minimal movement □ Flexed, some movement □ Stuck in extension, minimal movement □ Extended, some movement □ No limitation	□ Too much pronation □ Too much supination □ No limitation	□ Stuck in extension □ Some flexion/extension □ Stuck in flexion □ No limitation	□ Too much internal rotation □ Some internal rotation □ No limitation	□ Yes, describe □ No limitation	□ Bimanual: □ Back of palm □ Palm to palm □ Crossover palm to palm □ Hand to forearm/chest □ Trunk □ Other body part:
□ R ■ L	Grasp water bottle	NA	0	1	2	□ Scissor between the fingers □ Back of thumb and finger (fingers bending to thumb) □ Thumb in palm □ Pinch	□ Stuck in flexion, minimal movement □ Flexed, some movement □ Stuck in extension, minimal movement □ Extended, some movement □ No limitation	□ Too much pronation □ Too much supination □ No limitation	□ Stuck in extension □ Some flexion/extension □ Stuck in flexion □ No limitation	□ Too much internal rotation □ Some internal rotation □ No limitation	□ Yes, describe □ No limitation	□ Bimanual: □ Back of palm □ Palm to palm □ Crossover palm to palm □ Hand to forearm/chest □ Trunk □ Other body part:
□ R □ L	Bring it to mouth	NA	0	1	2	□ Scissor between the fingers □ Back of thumb and finger (fingers bending to thumb) □ Thumb in palm □ Pinch	□ Stuck in flexion, minimal movement □ Flexed, some movement □ Stuck in extension, minimal movement □ Extended, some movement □ No limitation	□ Too much pronation □ Too much supination □ No limitation	□ Stuck in extension □ Some flexion/extension □ Stuck in flexion □ No limitation	□ Too much internal rotation □ Some internal rotation □ No limitation	□ Yes, describe □ No limitation	□ Bimanual: □ Back of palm □ Palm to palm □ Crossover palm to palm □ Hand to forearm/chest □ Neck flexion □ Wrist flexion to reach □ Supination □ Passive elbow flexion □ Other body part:

Remarks: please describe any compensatory movements: use opposite arm, a particular starting position (standing versus sitting or assistive device such as sock aid, larger zipper pull, etc.) that the child may use to complete the task.

Describe pinch/grasp pattern (symmetrical or asymmetrical):

Describe any deviation (ulnar or radial) if present:

Describe any congenital anomaly (camptodactyly, pterygium)

14. Open the jar, pour out beads, pick up bead and string 3 beads onto string, and close the jar.

Arm	Subtask	Task Completion				Subtask Analysis of Joint Motion and Position						
						Prehension	Limitation				Adaptive Equipment/ Physical Assistance	Compensatory Movements
							Wrist	Forearm	Elbow	Shoulder		
☐ R ☐ L	Stabilizes jar to open	NA	0	1	2	☐ Scissor between the fingers ☐ Back of thumb and finger (fingers bending to thumb) ☐ Thumb in palm ☐ Pinch	☐ Stuck in flexion, minimal movement ☐ Flexed, some movement ☐ Stuck in extension, minimal movement ☐ Extended, some movement ☐ No limitation	☐ Too much pronation ☐ Too much supination ☐ No limitation	☐ Stuck in extension ☐ Some flexion/extension ☐ Stuck in flexion ☐ No limitation	☐ Too much internal rotation ☐ Some internal rotation ☐ No limitation	☐ Yes, describe ☐ No limitation	☐ Bimanual: ☐ Back of palm ☐ Palm to palm ☐ Crossover palm to palm ☐ Hand to forearm/chest ☐ Trunk ☐ Stabilize using surface or body ☐ Arm swing ☐ Other body part:
☐ R ☐ L	Open jar	NA	0	1	2	☐ Scissor between the fingers ☐ Back of thumb and finger (fingers bending to thumb) ☐ Thumb in palm ☐ Pinch	☐ Stuck in flexion, minimal movement ☐ Flexed, some movement ☐ Stuck in extension, minimal movement ☐ Extended, some movement ☐ No limitation	☐ Too much pronation ☐ Too much supination ☐ No limitation	☐ Stuck in extension ☐ Some flexion/extension ☐ Stuck in flexion ☐ No limitation	☐ Too much internal rotation ☐ Some internal rotation ☐ No limitation	☐ Yes, describe ☐ No limitation	☐ Bimanual: ☐ Back of palm ☐ Palm to palm ☐ Crossover palm to palm ☐ Hand to forearm/chest ☐ Trunk ☐ Stabilize using surface or body ☐ Arm swing ☐ Other body part:
☐ R ☐ L	Pour out	NA	0	1	2	☐ Scissor between the fingers ☐ Back of thumb and finger (fingers bending to thumb) ☐ Thumb in palm ☐ Pinch	☐ Stuck in flexion, minimal movement ☐ Flexed, some movement ☐ Stuck in extension, minimal movement ☐ Extended, some movement ☐ No limitation	☐ Too much pronation ☐ Too much supination ☐ No limitation	☐ Stuck in extension ☐ Some flexion/extension ☐ Stuck in flexion ☐ No limitation	☐ Too much internal rotation ☐ Some internal rotation ☐ No limitation	☐ Yes, describe ☐ No limitation	☐ Bimanual: ☐ Back of palm ☐ Palm to palm ☐ Crossover palm to palm ☐ Hand to forearm/chest ☐ Neck flexion ☐ Trunk ☐ Stabilize using surface or body ☐ Arm swing ☐ Other body part:

☐ R ☐ L	Hold string	NA	0	1	2						□ Pipe Cleaner	□ Stabilize using surface or body □ Trunk □ Other body parts
☐ R ☐ L	Pick up bead	NA	0	1	2	□ Scissor between the fingers □ Back of thumb and finger (fingers bending to thumb) □ Thumb in palm □ Pinch	□ Stuck in flexion, minimal movement □ Flexed, some movement □ Stuck in extension, minimal movement □ Extended, some movement □ No limitation	□ Too much pronation □ Too much supination □ No limitation	□ Stuck in extension □ Some flexion/extension □ Stuck in flexion □ No limitation	□ Too much internal rotation □ Some internal rotation □ No limitation	□ Yes, describe □ No limitation	□ Bimanual: □ Back of palm □ Palm to palm □ Crossover palm to palm □ Hand to forearm/chest □ Neck flexion □ Trunk □ Stabilize using surface or body □ Arm swing □ Other body part:
■ R ■ L	String 3 beads	NA	0	1	2						□ Pipe Cleaner	□ Work surface □ Trunk □ Other body parts

Remarks: please describe any compensatory movements: use opposite arm, a particular starting position (standing versus sitting or assistive device such as sock aid, larger zipper pull, etc.) that the child may use to complete the task.

Describe pinch/grasp pattern (symmetrical or asymmetrical):

Describe any deviation (ulnar or radial) if present:

Describe any congenital anomaly (camptodactyly, pterygium)

15. Pick up the marker, draw a circle or lines or write your name on this piece of paper with the marker, then cut or snip the paper (2-3 cuts in paper) using the scissors.

Arm	Subtask	Task Completion				Subtask Analysis of Joint Motion and Position						
						Prehension	Limitation				Adaptive Equipment/ Physical Assistance	Compensatory Movements
							Wrist	Forearm	Elbow	Shoulder		
☐ R ☐ L	Pick up the marker	NA	0	1	2	☐ Scissor between the fingers ☐ Back of thumb and finger (fingers bending to thumb) ☐ Thumb in palm ☐ Pinch	☐ Stuck in flexion, minimal movement ☐ Flexed, some movement ☐ Stuck in extension, minimal movement ☐ Extended, some movement ☐ No limitation	☐ Too much pronation ☐ Too much supination ☐ No limitation	☐ Stuck in extension ☐ Some flexion/extension ☐ Stuck in flexion ☐ No limitation	☐ Too much internal rotation ☐ Some internal rotation ☐ No limitation	☐ Yes, describe ☐ No limitation	☐ Bimanual: ☐ Back of palm ☐ Palm to palm ☐ Crossover palm to palm ☐ Hand to forearm/chest ☐ Neck flexion ☐ Trunk ☐ Stabilize using surface or body ☐ Arm swing ☐ Other body part:
☐ R ☐ L	Write / draw something	NA	0	1	2	☐ Palmar supinate ☐ Digital pronate ☐ Scissor between the fingers ☐ Back of thumb and finger (fingers bending to thumb) ☐ Thumb in palm ☐ Tripod grasp	☐ Stuck in flexion, minimal movement ☐ Flexed, some movement ☐ Stuck in extension, minimal movement ☐ Extended, some movement ☐ No limitation	☐ Too much pronation ☐ Too much supination ☐ No limitation	☐ Stuck in extension ☐ Some flexion/extension ☐ Stuck in flexion ☐ No limitation	☐ Too much internal rotation ☐ Some internal rotation ☐ No limitation	☐ Yes, describe ☐ No limitation	☐ Bimanual: ☐ Back of palm ☐ Palm to palm ☐ Crossover palm to palm ☐ Hand to forearm/chest ☐ Neck flexion ☐ Trunk ☐ Stabilize using surface or body ☐ Arm swing ☐ Other body part:
☐ R ☐ L	Stabilize paper to cut	NA	0	1	2							☐ Bimanual: ☐ Back of palm ☐ Palm to palm ☐ Crossover palm to palm ☐ Hand to forearm/chest ☐ Neck flexion ☐ Trunk ☐ Stabilize using surface or body ☐ Arm swing ☐ Other body part:

☐ R ☐ L	Cut paper	NA	0	1	2	☐ Thumb and fingers ☐ Fingers only	☐ Stuck in flexion, minimal movement ☐ Flexed, some movement ☐ Stuck in extension, minimal movement ☐ Extended, some movement No limitation	☐ Too much pronation ☐ Too much supination ☐ No limitation	☐ Stuck in extension ☐ Some flexion/extension ☐ Stuck in flexion No limitation	☐ Too much internal rotation ☐ Some internal rotation ☐ No limitation	☐ Yes, describe ☐ No limitation	☐ Bimanual: ☐ Back of palm ☐ Palm to palm ☐ Crossover palm to palm ☐ Hand to forearm/chest ☐ Neck flexion ☐ Trunk ☐ Stabilize using surface or body ☐ Arm swing ☐ Other body part:

Remarks: please describe any compensatory movements: use opposite arm, a particular starting position (standing versus sitting or assistive device such as sock aid, larger zipper pull, etc.) that the child may use to complete the task.

Describe pinch/grasp pattern (symmetrical or asymmetrical):

Describe any deviation (ulnar or radial) if present:

Describe any congenital anomaly (camptodactyly, pterygium)

16. Put on this vest, attach the zipper, and pull the zipper all the way up and then all the way back down.

Arm	Subtask	Task Completion				Subtask Analysis of Joint Motion and Position						
						Prehension	Limitation				Adaptive Equipment/ Physical Assistance	Compensatory Movements
							Wrist	Forearm	Elbow	Shoulder		
☐ R ☐ L	Put on item of clothing	NA	0	1	2						☐ Bigger zipper pull ☐ Yes, describe ☐ No limitation	☐ Unilateral ☐ Stabilize using surface or body ☐ Arm swing ☐ Other body part:
☐ R ☐ L	Stabilize zipper	NA	0	1	2	☐ Scissor between the fingers ☐ Back of thumb and finger (fingers bending to thumb) ☐ Thumb in palm ☐ Pinch	☐ Stuck in flexion, minimal movement ☐ Flexed, some movement ☐ Stuck in extension, minimal movement ☐ Extended, some movement ☐ No limitation	☐ Too much pronation ☐ Too much supination ☐ No limitation	☐ Stuck in extension ☐ Some flexion/extension ☐ Stuck in flexion ☐ No limitation	☐ Too much internal rotation ☐ Some internal rotation ☐ No limitation	☐ Bigger zipper pull ☐ Yes, describe ☐ No limitation	☐ Bimanual: ☐ Back of palm ☐ Palm to palm ☐ Crossover palm to palm ☐ Hand to forearm/chest ☐ Neck flexion ☐ Trunk ☐ Stabilize using surface or body ☐ Arm swing ☐ Other body part:
☐ R ☐ L	Attach zipper	NA	0	1	2	☐ Scissor between the fingers ☐ Back of thumb and finger (fingers bending to thumb) ☐ Thumb in palm ☐ Pinch	☐ Stuck in flexion, minimal movement ☐ Flexed, some movement ☐ Stuck in extension, minimal movement ☐ Extended, some movement No limitation	☐ Too much pronation ☐ Too much supination ☐ No limitation	☐ Stuck in extension ☐ Some flexion/extension ☐ Stuck in flexion No limitation	☐ Too much internal rotation ☐ Some internal rotation ☐ No limitation	☐ Bigger zipper pull ☐ Yes, describe ☐ No limitation	☐ Bimanual: ☐ Back of palm ☐ Palm to palm ☐ Crossover palm to palm ☐ Hand to forearm/chest ☐ Neck flexion ☐ Trunk ☐ Stabilize using surface or body ☐ Arm swing Other body part:
☐ R ☐ L	Pull zipper halfway to chest	NA	0	1	2	☐ Scissor between the fingers ☐ Back of thumb and finger (fingers bending to thumb) ☐ Thumb in palm	☐ Stuck in flexion, minimal movement ☐ Flexed, some movement ☐ Stuck in extension,	☐ Too much pronation ☐ Too much supination ☐ No limitation	☐ Stuck in extension ☐ Some flexion/extension ☐ Stuck in flexion No limitation	☐ Too much internal rotation ☐ Some internal rotation	☐ Bigger zipper pull ☐ Yes, describe	☐ Bimanual: ☐ Back of palm ☐ Palm to palm ☐ Crossover palm to palm

						□ Pinch	minimal movement □ Extended, some movement □ No limitation			□ No limitation	□ No limitation	□ Hand to forearm/chest □ Neck flexion □ Trunk □ Stabilize using surface or body □ Arm swing □ Other body part:
☐ R ☐ L	Pull zipper up to chest	NA	0	1	2	□ Scissor between the fingers □ Back of thumb and finger (fingers bending to thumb) □ Thumb in palm □ Pinch	□ Stuck in flexion, minimal movement □ Flexed, some movement □ Stuck in extension, minimal movement □ Extended, some movement □ No limitation	□ Too much pronation □ Too much supination □ No limitation	□ Stuck in extension □ Some flexion/extension □ Stuck in flexion □ No limitation	□ Too much internal rotation □ Some internal rotation □ No limitation	□ Bigger zipper pull □ Yes, describe □ No limitation	□ Bimanual: □ Back of palm □ Palm to palm □ Crossover palm to palm □ Hand to forearm/chest □ Neck flexion □ Trunk □ Stabilize using surface or body □ Arm swing □ Other body part:

Remarks: please describe any compensatory movements: use opposite arm, a particular starting position (standing versus sitting or assistive device such as sock aid, larger zipper pull, etc.) that the child may use to complete the task.

Describe pinch/grasp pattern (symmetrical or asymmetrical):

Describe any deviation (ulnar or radial) if present:

Describe any congenital anomaly (camptodactyly, pterygium)

17. Hold the dodgeball reach the sky and throw it towards me.

Arm	Subtask	Task Completion				Subtask Analysis of Joint Motion and Position							
						Prehension	Limitation					Adaptive Equipment/ Physical Assistance	Compensatory Movements
							Wrist	Forearm	Elbow	Shoulder	Shoulder		
■ R □ L	Throw ball (overhead no compensation=2, overhead with compensation =1, cannot throw overhead=0)	NA	0	1	2	□ Palm to palm □ Crossover palm to palm □ Asymmetrical grasp	□ Stuck in flexion, minimal movement □ Flexed, some movement □ Stuck in extension, minimal movement □ Extended, some movement □ No limitation	□ Too much pronation □ Too much supination □ No limitation	□ Stuck in extension □ Some flexion/extension □ Stuck in flexion □ No limitation	□ Too much internal rotation □ Some internal rotation □ No limitation	□ No/Minimal flexion □ Some flexion □ No limitation (has more than 90 degree)	□ Yes, describe □ No limitation	□ Bimanual: □ Back of palm □ Palm to palm □ Crossover palm to palm □ Hand to forearm/chest □ Neck flexion □ Trunk □ Stabilize using surface or body □ Arm swing □ Other body part:
□ R ■ L	Throw ball (overhead no compensation=2, overhead with compensation =1, cannot throw overhead=0)	NA	0	1	2	□ Palm to palm □ Crossover palm to palm □ Asymmetrical grasp	□ Stuck in flexion, minimal movement □ Flexed, some movement □ Stuck in extension, minimal movement □ Extended, some movement □ No limitation	□ Too much pronation □ Too much supination □ No limitation	□ Stuck in extension □ Some flexion/extension □ Stuck in flexion □ No limitation	□ Too much internal rotation □ Some internal rotation □ No limitation	□ No/Minimal flexion □ Some flexion □ No limitation (has more than 90 degree)	□ Yes, describe □ No limitation	□ Bimanual: □ Back of palm □ Palm to palm □ Crossover palm to palm □ Hand to forearm/chest □ Neck flexion □ Trunk □ Stabilize using surface or body □ Arm swing □ Other body part:

Remarks: please describe any compensatory movements: use opposite arm, a particular starting position (standing versus sitting or assistive device such as sock aid, larger zipper pull, etc.) that the child may use to complete the task.

Describe pinch/grasp pattern (symmetrical or asymmetrical):

Describe any deviation (ulnar or radial) if present:

Describe any congenital anomaly (camptodactyly, pterygium)

18. Pull down the shorts, reach your bottom from the back while holding 2 squares of toilet paper, also reach between your legs from the front while holding 2 squares of toilet paper, and pull the shorts back up.

Arm	Subtask	Task Completion				Subtask Analysis of Joint Motion and Position						
						Prehension	Limitation				Adaptive Equipment/ Physical Assistance	Compensatory Movements
							Wrist	Forearm	Elbow	Shoulder		
☐ R ☐ L	Pull down shorts	NA	0	1	2						☐ Yes, describe ☐ No limitation	☐ Trunk ☐ Stabilize using surface or body ☐ Arm swing ☐ Other body part:
☐ R ☐ L	Reach buttocks area while holding TP [back]	NA	0	1	2	☐ Scissor between the fingers ☐ Back of thumb and finger (fingers bending to thumb) ☐ Thumb in palm ☐ Pinch	☐ Stuck in flexion, minimal movement ☐ Flexed, some movement ☐ Stuck in extension, minimal movement ☐ Extended, some movement ☐ No limitation	☐ Too much pronation ☐ Too much supination ☐ No limitation	☐ Stuck in extension ☐ Some flexion/extension ☐ Stuck in flexion ☐ No limitation	☐ Too much internal rotation ☐ Some internal rotation ☐ No limitation	☐ Yes, describe ☐ No limitation	☐ Trunk ☐ Stabilize using surface or body ☐ Arm swing ☐ Other body part:
☐ R ☐ L	Reach between legs while holding TP [front]	NA	0	1	2	☐ Scissor between the fingers ☐ Back of thumb and finger (fingers bending to thumb) ☐ Thumb in palm ☐ Pinch	☐ Stuck in flexion, minimal movement ☐ Flexed, some movement ☐ Stuck in extension, minimal movement ☐ Extended, some movement ☐ No limitation	☐ Too much pronation ☐ Too much supination ☐ No limitation	☐ Stuck in extension ☐ Some flexion/extension ☐ Stuck in flexion ☐ No limitation	☐ Too much internal rotation ☐ Some internal rotation ☐ No limitation	☐ Yes, describe ☐ No limitation	☐ Trunk ☐ Stabilize using surface or body ☐ Arm swing ☐ Other body part:
☐ R ☐ L	Pull up shorts	NA	0	1	2		☐ Stuck in flexion, minimal movement ☐ Flexed, some movement ☐ Stuck in extension, minimal movement ☐ Extended, some movement ☐ No limitation	☐ Too much pronation ☐ Too much supination ☐ No limitation	☐ Stuck in extension ☐ Some flexion/extension ☐ Stuck in flexion ☐ No limitation		☐ Yes, describe ☐ No limitation	☐ Trunk ☐ Stabilize using surface or body ☐ Arm swing ☐ Other body part:

Remarks: please describe any compensatory movements: use opposite arm, a particular starting position (standing versus sitting or assistive device such as sock aid, larger zipper pull, etc.) that the child may use to complete the task.

Describe pinch/grasp pattern (symmetrical or asymmetrical):

Describe any deviation (ulnar or radial) if present:

Describe any congenital anomaly (camptodactyly, pterygium)