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Functioning of Young Individuals with Upper Limb Reduction Deficiencies

Golea-Vasluian, Ecaterina

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Chapter 8

Discussion and implications

This is me, Amy! is an inspirational book about an eleven-year-old girl with a reduction deficiency of her left arm.¹ The book gives an insightful view into Amy's life experiences with an upper limb reduction deficiency (ULRD) and invites children and parents to engage together in the challenging journey of young individuals with ULRD and to communicate and find solutions to the difficulties encountered. For example, because she was unable to play the saxophone with just one hand, Amy chose to play the trumpet.

This thesis joins the journey of children, their parents, and healthcare professionals towards enabling activities and participation of young individuals with ULRD, and it aims to

- (a) determine the current numbers of children with upper and lower limb deficiencies in the Netherlands,
- (b) identify possible difficulties in functioning and enabling solutions,
- (c) assess the role of and satisfaction with some of the used solutions (prostheses and adaptive devices [ADs]) and satisfaction with rehabilitation care, and
- (d) provide an appropriate functional test to evaluate hand function of young individuals with ULRD in activities of daily living (ADL).

This thesis produces valuable information for healthcare professionals, children with ULRD and their parents, and researchers.

Main results

ICF data about body structures and functions, environmental and personal factors, and activities and participation of young individuals with ULRD are reported in the thesis (see overview of main results, Table 1). In short, the birth prevalence for ULRD was 5 per 10,000 births during 1981–2010 in the northern Netherlands (*chapter 2*). Due to the affected body structure, young individuals with ULRD are confronted with stares and adverse reactions from people in their environment or difficulties in some ADL. To avoid stares and adverse reactions, young individuals with ULRD mainly use prostheses (*chapter 3*). To facilitate difficult ADL, ADs are used preferentially over prostheses for self-care, mobility, and leisure activities (*chapter 4*). Two functional tests evaluating function of the affected upper limb are described: SHAP and the adapted version for pediatric use, SHAP-C. When applied to novice prosthetic users, SHAP scores were impacted by learning effects (*chapter 5*). For SHAP-C, intra-rater reliability was better than inter-rater reliability (*chapter 6*). In the last chapter,

chapter 7, an alternative SHAP scoring system is proposed; this new system solves issues with the current one such as cumbersome formulas, unclear influence of contributing tasks, and standardization of patient times using standard deviations of unimpaired persons.

Broader perspective of results

ULRD–epidemiology, functioning, prostheses, and adaptive devices

The birth prevalence for congenital limb defects in the northern Netherlands for a 30-year period (1981–2010) was derived from the European Surveillance of Congenital Anomalies in the northern Netherlands database (EUROCAT-NNL). EUROCAT-NLL is part of the EUROCAT network of national registries that collect, manage, and analyze data on birth defects.^{2,3} EUROCAT’s main purpose is to provide estimates of the occurrence of birth defects and identify timely trends, associations between different defects, and possible causes for the birth defects.

Compared to other European countries that are part of EUROCAT, the calculated birth prevalence for CLD in the Netherlands (20.0/10,000, including polydactyly, syndactyly, and reduction defects only) was similar to Italy (Emilia-Romagna, 18.0 and Tuscany, 18.6) and Denmark (Odense, 22.2/10,000) for the same period of time (*chapter 2*). However, there is only limited comparability to international data due to variability in definitions for the inclusion or classification of CLD in different countries. The reported birth prevalence for reduction defects in Canada varied from 3.3 to a peak of 8.1 (1966–1970) and decreased afterwards to 5.0 (1975); in the USA, it decreased from 6.6 to 4.8/10,000 (1968–1993).⁴⁻⁶ The Dutch birth prevalence for reduction defects of 6.9/10,000 births is comparable with Belgium (EUROCAT registry Hainaut, 7.3/10,000). The birth prevalence for ULRD (5/10,000) is similar to Finland (5.6/10,000, 1995–2005),⁷ but higher than Sweden (3.9/10,000, 1997–2007).⁸ However, the studies in Finland and Sweden used Swanson’s classification, which is different from that used in the present study;⁹ this restricts direct comparability with the Dutch birth prevalence. Although ULRD are rare, young individuals with ULRD require attention, as throughout growth, they may encounter difficulties in functioning as described later in the current thesis.

The results of this thesis confirmed that, in general, young individuals with ULRD function well psychosocially and physically, irrespective of the use of prostheses or adaptive

devices (*chapters 3 and 4*). Psychosocial functioning of children with ULRD was comparable with the norms of unimpaired children.¹⁰ Studies evaluating the impact of prostheses on the physical functioning of young individuals with ULRD found comparable performance in ADL with or without prostheses.^{11,12} Performing well does not necessarily mean that young individuals with ULRD experience no difficulty in ADL. They do have difficulties in ADL that require both hands,¹³ for instance for self-care (using cutlery or tying shoelaces), mobility (riding a bicycle), or in recreation and leisure activities (playing sports or playing a musical instrument; *chapter 4*).

Solutions to enable functioning

The use of compensatory or adapted movements by young individuals with ULRD has been described as a solution to perform activities and facilitate participation.^{14,15} Our results positioned the use of unaffected and affected upper limbs and other body parts as preferred solutions for ADL (*chapter 4*). Along with impaired upper limb structure, the compensatory movements trigger abnormal body movements that draw the attention of people in the environment. Staring and adverse reactions distress young individuals with ULRD and become more bothersome during transitional periods, such as new environments or life phases (e.g., puberty).^{14,16,17} Confronted with personal factors such as self-identity, sexuality, and careers,¹⁴ they wish for solutions that would allow for a normal appearance. Thus, prostheses are worn for such purposes, including cosmetic enhancements and normal appearance (*chapter 3*). We and others have reported activity-specific use of the prostheses for improvement of physical functioning, for instance, lifting heavy objects or participating in sports.^{12,18} Nevertheless, every child experiences ULRD in his or her own way.¹ Many accept the ULRD and do not need cosmetic improvement; others see prostheses as highlighting the ULRD or are overwhelmed by the technical and functional limitations of prostheses (*chapter 3*). Prostheses are then rejected.

Difficulties in ADL, however, continue to persist, leading to a search for alternative solutions. The adaptive devices seem to be used for specific ADL (e.g. using cutlery, riding a bicycle, or playing a musical instrument) and to provide the needed functionality (*chapters 3 and 4*).

ULRD–functional tests: SHAP and SHAP-C

The Upper Limb Prosthetic Outcomes group suggested investigating the psychometric properties of SHAP in prosthetic users.^{19,20} As a result, we focused on learning effects in novice users, since SHAP is often used to monitor training of novice prosthetic users. Our study found learning effects in 24 unimpaired participants that used a prosthetic simulator (myoelectric prosthesis mountable on the forearm). Contrary to our findings, another study found no learning effects in SHAP; however, that study included only one participant who was experienced with SHAP.²¹ Another issue was identified with the time limits for each SHAP task. Tasks such as picking up coins, cutting food, or rotating a screw could be performed, but it took a longer time. According to our interpretation of formulas used to calculate SHAP scores from task times, exceeding the task time limit affects scores. Sometimes, scores are influenced considerably if many tasks are affected. After having consulted an experienced statistician and following unsuccessful attempts to obtain a detailed description from the developers of SHAP, difficulties in clearly understanding the SHAP scoring system urged us to suggest a new simplified scoring system (*chapter 6*). Due to the cumbersome calculations and the lack of transparency, it is difficult to trust SHAP scores.

During and after the reliability tests of the adapted version of SHAP for children, SHAP-C, some issues were observed. Adaptation of SHAP for pediatric use included downsizing the objects used for measurements, starting-stopping moments were modified, and the assessors were used to collect data. The SHAP-C protocol was kept as close as possible to the original SHAP protocol. However, either due to the natural variability in pediatric performance or the influence of the assessor (response time), inter-rater reliability was poor. Therefore, SHAP-C should be used with caution when more assessors are used; further improvements are required to provide clinicians with a reliable SHAP-C.

Suggestions for rehabilitation and future research

Comparing the birth prevalence in the Netherlands with international values was rather difficult due to differences in registration methods (e.g., inclusion criteria, assessment method, or classifications used). Using a unique registration system would make comparison of international data more reliable. Furthermore, combining data from sources with the same

registration methods would allow for larger sample sizes and, thus, more trustworthy estimates. More comprehensive classifications of congenital birth defects are yet to be developed since issues with the existing ones have been reported (e.g., Swanson classification);^{7,8} we have also observed these issues.

The results of this thesis promote the use of prostheses and adaptive devices in young individuals with ULRD needing psychosocial and physical rehabilitation for functioning. The prostheses seem to enable better cosmesis in young individuals with ULRD that wish for a normal appearance. The adaptive devices facilitate physical functioning in ADL, independence in activities, and participation. Cosmesis and independence seem to contribute to the concept of “normality.”²² When done according to these components of normality, normal appearance, and independence, rehabilitating young individuals with ULRD enables them to have experiences like their peers and to undergo social integration. Prosthetic manufacturers and rehabilitation professionals should therefore focus on providing young individuals with ULRD with desired cosmesis or desired independence. Insurance companies are often reluctant regarding the compensation of costs for cosmetic prostheses. Our findings showed that there is a clear need for such prostheses, especially during certain phases of life (e.g., puberty). The adaptive devices are valuable alternatives to prostheses and should also be included in standard rehabilitation care as they account for the drawbacks of prostheses (increased weight and difficulties with manipulation). For underdeveloped countries, introduction of adaptive devices as alternatives to prostheses may be particularly beneficial regarding costs and successful rehabilitation of young individuals with ULRD. Experienced technicians should share knowledge about the manufacturing aspects of adaptive devices at an international level.

Conversely, the preferred use of the affected and unaffected upper limbs and other body parts is not surprising. Persons with congenital reduction deficiencies use the affected upper limb more than persons with acquired amputation.²³ Consequently, rehabilitation professionals must be careful when applying knowledge about prosthetic preferences of persons with upper limb amputation to rehabilitation of functioning in young individuals with ULRD.

Furthermore, switching from the idea that “healthcare professionals know what is best for the patient” to a patient-centered approach is necessary and more efficient. Identifying the individual needs of young individuals and their parents regarding functioning with ULRD,

finding suitable enabling solutions, and balancing patients' expectations regarding the real possibilities of solutions are recommended for rehabilitation. With or without the use of adaptive devices or prostheses, growing up dexterous and with a positive self-image will help young individuals with ULRD integrate into society.

Young individuals with ULRD make efforts to combat stares and adverse reactions from people in their environment. Why not change the views of people? Maybe common efforts of healthcare providers, teachers, parents, or even social media would improve the quality of life in young individuals with ULRD and people with impairments in general. A first step would be to increase awareness in the general population about the consequences of the social stigmata that people with impairments experience. Promoting children's stories such as the one about Amy in the form of books (cartoons, movies, or social media) to a broader public,¹ including young individuals with ULRD in TV shows, or stimulating active participation of young individuals with ULRD in group activities in school provide a few examples of such initiatives.

SHAP is a promising functional test that has several important advantages for clinicians and researchers: a comprehensive overview of the functionality of six hand grips and general functionality that are assessed objectively in < 30 minutes. Nevertheless, more research is needed on the psychometric properties, including learning effects, in experienced prosthetic users. In novice prosthetic users, the occurrence of learning effects imposes caution when using SHAP to monitor training outcomes. Time limits and their influence, when exceeded, on SHAP functional scores need further consideration in case of prosthetic users. In novice users, time limits were often exceeded.

Further studies need to consider the observed issues with SHAP-C, including fluctuating motivation, reaction time, influence of the assessor, or finding an alternative and suitable data collection method. Our explorative study may be a first step towards the development of a reliable SHAP-C.

Strengths and limitations

Research questions have been answered with the use of different study designs: retrospective cohort, qualitative, cross-sectional, longitudinal, and repeated measures studies with corresponding advantages and disadvantages.

In *chapter 2*, the epidemiological data covered a large period, over 30 years. The novelty of the studies in *chapters 3 and 4* was the inclusion of the younger children and adolescents with ULRD along with their parents and healthcare professionals. Their opinions provided a complete picture about the experiences and needs of young individuals with ULRD. Using online focus group interviews allowed for a more open interaction between anonymous participants, as well as time and cost-efficient collection of data (*chapter 3*). The study in *chapter 4* is the first one that addressed the use and satisfaction with adaptive devices compared to prostheses in young individuals with ULRD. The studies about SHAP and SHAP-C on psychometric aspects (*chapters 5, 6, and 7*) contribute knowledge for further improvement of the tests.

Information bias may have affected some of our studies requiring answers about previous events (*chapters 2, 3, and 4*). The EUROCAT database is based on the questionnaire completed by parents or guardians of children with birth defects. Luckily, EUROCAT has a well-established system for notifying cases promptly after birth, usually by the hospitals and general practitioners.²⁴ Parents are then contacted and asked to provide characteristics of their child and themselves, thus minimizing recall bias. Furthermore, hospital files and results of investigations are made available for EUROCAT to double-check the information. In the qualitative study, some of the participants could not comment on the perceived rehabilitation care because they did not remember it anymore.

In the study evaluating learning effects of SHAP tasks in novice prosthetic users, participants were unimpaired persons using a prosthetic simulator, not actual prosthetic users. Finding prosthetic users with the same baseline characteristics and no experience with SHAP is difficult. Prosthetic users also differ in their capabilities to use a prosthesis, which would create inequalities between participants.²⁵ Many prosthetic users are likely to be familiar with SHAP since the procedure is used in many Dutch rehabilitation centers, as well as worldwide. Including participants acquainted with SHAP would have biased detection of learning effects. Finding actual prosthetic users is often difficult for researchers. Therefore, using unimpaired

persons and a prosthetic simulator is advantageous, since larger samples could be included. Furthermore, research provided evidence that performances of prosthetic users and unimpaired persons using a simulator are comparable.²⁶

The questionnaires used in *chapter 4* were not validated for a pediatric population because it would have required validating the questionnaire in the same population that was included in the survey.

The response rate in our study using questionnaires to evaluate use of adaptive devices was not as high (43%) as desired. Decreasing trends in response rates have been reported for the last several years.^{27,28} Because they seem to perceive themselves as unimpaired and generally function well, young individuals with ULRD may not have been interested in participating.^{11,12,14}

Conclusion

This thesis addressed knowledge gaps on functioning of young individuals with ULRD; this was accomplished on different levels. The identified needs for using prostheses and adaptive devices were normal appearance and independence in ADL; both components were important for achieving social integration. Functional evaluation is a milestone step in rehabilitation of young individuals with ULRD. SHAP and the adapted version for children, SHAP-C, are functional tests that provide comprehensive data about the functional capabilities with the ULRD. The issues described in our studies with SHAP and SHAP-C should serve as a base for the prospective work needed to provide healthcare professionals with reliable tests.

Table 1. Main thesis findings structured under the ICF components

CHAPTER/STUDY	BODY STRUCTURES & FUNCTIONS	ACTIVITIES & PARTICIPATION	PERSONAL & ENVIRONMENTAL FACTORS
Chapter 2 Birth prevalence of congenital limb defects	Limb deficiencies 21.1/10,000 Reduction deficiencies 6.9 ULRD 5.0 Transversal ULRD 2.9 Isolated ULRD 2.6 Recognized condition ULRD 1.9		
Chapter 3 Prosthetic use and rehabilitation care	Wearing prostheses improved <i>cosmetic</i> appearance (complete body) and psychosocial functioning (<i>normality, social integration</i>).	Young individuals with ULRD were able to perform ADL with or without prostheses. ADL were facilitated by the use of prostheses (lifting heavy objects, some sports).	Young individuals with ULRD used prostheses to avoid stares and adverse reactions and to perform as peers and rejected prostheses due to their unnatural appearance or due to acceptance of ULRD. Prosthesis use or non-use was also influenced by parental involvement or technical (heavy, non-durable gloves) and functional limitations. Rehabilitation care was perceived as appropriate.
Chapter 4 ADs vs. prostheses, use/satisfaction/social adjustment	Physical and psychosocial functioning were facilitated by the use of ADs (<i>bimanual/peer-like functioning</i>).	Young individuals with ULRD preferred to use unaffected and affected upper limbs and other body parts to perform ADL. More than 50% of young individuals with ULRD reported difficulties in ADL. The use ADs facilitated self-care, mobility, recreation, and leisure ADL.	Young individuals with ULRD needed ADs for <i>independent and easy</i> performance in ADL. Young individuals' satisfaction with ADs > prostheses. Satisfaction with rehabilitation was high and comparable between ADs and prostheses. ADs, provided by rehabilitation teams or family, were regarded as inexpensive, light, and easy to manipulate.
CHAPTER/STUDY	BODY FUNCTIONS/ACTIVITIES & PARTICIPATION – measurement of ULRD function in ADL		
Chapter 5 SHAP, learning effects	SHAP is a measurement test evaluating upper limb functioning; it includes ADL tasks and generates individual hand grip and overall scores. SHAP functional scores may not represent the actual physical abilities with the prosthetic upper limb in novice users. SHAP tasks were affected by <i>learning effects</i> when performed repeatedly in novice prosthetic users using strategies to perform faster. Some SHAP tasks required more execution time, which caused exceeding the time limits of the corresponding tasks.		
Chapter 6 SHAP-C, reliability	Children were able to grasp all SHAP-C objects and perform all SHAP-C tasks with the unimpaired or prosthetic hand. <i>Intra-rater</i> reliability of SHAP-C was <i>better</i> than the <i>inter-rater</i> reliability in unimpaired children. Fluctuating motivation and assessors' reaction times possibly affected reliability. Timing the performance of SHAP-C tasks may not be appropriate for children.		
Chapter 7 SHAP, alternative scoring system	A <i>new and improved scoring system</i> was proposed for SHAP. The standardized task times used to calculate the functional scores are free of the influence of variability (standard deviation) in norms (unimpaired young adults). SHAP tasks have a better representation in the functional scores.		

Abbreviations and notations: ICF, International Classification of Functioning, Disability, and Health; ULRD, upper limb reduction deficiencies; AD, adaptive devices; ADL, activities of daily living; '>', higher than.

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Summary

Children with upper limb reduction deficiencies (ULRD) are born with a part of their limb(s) missing, which may cause different degrees of impairment. The impairment affects their physical and psychosocial functioning. Consequently, healthcare professionals mainly provide prostheses to enable functioning of young individuals (defined as ages 2–20 years) with ULRD. With or without prostheses, young individuals with ULRD have comparable functionality in daily activities. Along with lack of functional gain, being heavy, difficult to manipulate, or uncomfortable, prostheses are rejected by 35–45% of young individuals with ULRD. The direct reasons why young individuals with ULRD choose to use or not use a prosthesis are underexplored, nor is feedback from such young individuals available, regarding their rehabilitation care. The existence of alternatives to facilitate difficult activities such as adaptive devices is only briefly described in the literature. However, actual evaluation of the satisfaction and social adjustment with and use of adaptive devices has not been done in young individuals with ULRD. Questions yet to be answered include “Are young individuals with ULRD more satisfied with prostheses or adaptive devices?” or “Is there a difference between social adjustment with a prosthesis and an adaptive device?”

To make better informed choices regarding rehabilitating young individuals with ULRD, clinicians need to evaluate the actual performance with the affected limb. The Southampton Hand Assessment Procedure (SHAP) is a functional test with several advantages over other instruments: universal applicability in assessment of (un)impaired and prosthetic hands; functional scores for specific grips, allowing focus on improving grips with low scores; and fast and easy data collection, which can be completed in 20 minutes using a transportable kit with 26 tasks. The adult version of SHAP (no pediatric version exists) needs to be tested for psychometric properties in prosthetic users. To use SHAP in children, the objects and protocol must be adapted.

This thesis was structured in two main parts. The first part aims to provide updated epidemiologic data about young individuals with congenital limb defects, investigate the effectiveness and impact of prostheses and adaptive devices in young individuals with ULRD, while the second part aims to evaluate psychometric properties of SHAP in prosthetic users and of the adapted version for children (SHAP-C).

The ultimate goal was to provide extensive knowledge that will benefit young individuals with ULRD, researchers, and healthcare providers.

The International Classification of Functioning, Disability, and Health (ICF) has been used to conduct and structure the research in this thesis. The ICF is a model recommended by the World Health Organization to describe health and disability status. This thesis contributes knowledge about the ICF components regarding children with ULRD, including body functions and structures, activities and participation, and personal and environmental factors.

I. Young individuals with ULRD—epidemiology, functioning, prostheses, and adaptive devices

A detailed description of the Dutch population affected by limb defects is given as an introduction in *chapter 2*. In the Netherlands, the birth prevalence of children born with an affected body structure—congenital limb deficiencies (CLD)—was found to be 21.1 per 10,000 births for the period 1981–2010. Reduction deficiencies affected 6.9 per 10,000 births, of which upper limb ULRD affected 5 per 10,000. The ULRD were mostly isolated affections (not part of a syndrome or other recognized condition).

Chapters 3 and 4 present qualitative and quantitative knowledge about components of ICF functioning, including body functions, activities, and personal and environmental factors. The young individuals with ULRD reported having difficulties in activities of daily living for which they preferentially used their affected and unaffected upper limbs and other body parts. When experienced difficulty in many activities, they opted for adaptive devices that were meant to improve functionality. Prostheses were much less preferred. Adaptive devices were used to facilitate activities of self-care, mobility, recreation, and leisure.

In online focus group interviews, young individuals with below-elbow ULRD, their parents, and healthcare professionals shared their opinions about reasons to use or not use prostheses and about rehabilitation care (*chapter 3*). The main reason for wearing a prosthesis was cosmesis, as the prosthesis helped to decrease stares, adverse reactions, and facilitated fitting into the environment. Among non-users, acceptance of the affected arm and avoiding the unnatural cosmesis of a prosthesis were reasons for not using one. The prostheses were regarded as useful accessories for specific activities such as lifting heavy objects or participating in sports. However, many of the young individuals with ULRD were able to perform most activities without a prosthesis. Commonly mentioned reasons for not wearing a prosthesis were increased weight, technical limitations, and difficulties with manipulating the prosthesis.

The role of environmental factors, including the parents, in prosthetic use is highlighted in *chapter 3*. Some parents wished to see their child “more complete” and chose a prosthesis; others did not believe a prosthesis would improve their child’s function and chose not to use one. Satisfaction with adaptive devices was significantly higher than satisfaction with prostheses (*chapter 4*). Young individuals with ULRD were more satisfied with the features of the adaptive devices, including lightweight and easy manipulation.

Rehabilitation care seems to meet the expectations of young individuals with ULRD and their parents. They reported their experience with rehabilitation care as appropriate (*chapter 3*) and were satisfied with it, regardless of the type of solutions used (prostheses or adaptive devices, *chapter 4*). Patient-centered care is suggested as a desirable approach in rehabilitation, and peer-contact was found to be important for exchanging information, emotional support, and coping with ULRD.

II. ULRD–functional tests: SHAP and SHAP-C

Chapters 5 and 6 report research on the psychometric properties of a functional test evaluating body functions of the affected arm while performing activities of daily living. The tasks of SHAP were impacted by learning effects when applied repeatedly in novice prosthetic users (unimpaired persons using a prosthetic simulator, *chapter 5*). A warm-up decrement in performance was also present; that is, in a set of two sessions, performance times in the first session were longer than those in the second, indicating poorer functionality in the first session. Novice users exceeded time limits imposed for each task on several occasions; this had repercussions on the functional scores for the hand grips and the overall score (index of function). Hence, clinicians should bear in mind these factors influencing prosthetic performance in novice users.

The first steps to adapt SHAP for children (SHAP-C) are presented in *chapter 6*. The downsized objects were appropriate for a pediatric hand or a prosthetic hand, and all unimpaired children were able to perform the SHAP-C tasks, which included the same tasks as in SHAP. The intra-rater reliability, evaluated with repeatability coefficients, was relatively good in comparison with inter-rater reliability. Possible issues such as pediatric natural variability in performance, fluctuating motivation, and assessors’ reaction times need to be accounted for in further studies on SHAP-C.

In *chapter 7*, a new, simple, and straightforward scoring system was proposed to calculate SHAP functional scores. In the new scoring system, the separate tasks have a different relation to the norms. In the original SHAP, the task times are standardized using z-scores and variability (standard deviations) of the norms derived from unimpaired subjects. Since the prosthetic users may have different standard deviations than those of unimpaired persons, in our study, the standardization of task times was free of the variability in norms from unimpaired persons. Furthermore, future research can determine how the influence of tasks on the functional score can be weighted. This means that the tasks/the hand grips that are more commonly used in daily performance will have a larger contribution to the functional scores. In SHAP, the tasks have a fixed contribution to the functional scores, and exceeding a time limit for a task may affect the functional scores to a greater degree.

The last chapter, *chapter 8*, provides a general discussion about the applicability of the main results for healthcare practice and future research. The use of prostheses and adaptive devices has an impact on the physical and psychosocial functioning and improves the normality of children with ULRD. The normality components, cosmesis and independence, should be addressed by rehabilitation professionals working with young individuals with ULRD, since normality enables social integration. Consideration of the identified issues with SHAP, learning effects, and with SHAP-C, poor inter-rater reliability and further research/development of the corresponding instruments are advised for clinicians and researchers.

Samenvatting

Kinderen met reductiedefecten van de armen (Upper Limb Reduction Deficiencies, hierna genoemd ULRD) hebben een aangeboren afwijking waarbij één of beide armen niet volledig zijn ontwikkeld. Een ULRD kan in meer of mindere mate tot een beperking leiden. Deze beperking kan invloed hebben op het fysieke en psychosociale functioneren van de kinderen. Om deze beperkingen te reduceren, adviseren behandelaars in de gezondheidszorg soms prothesen aan de kinderen en jongeren (2-20 jaar). Kinderen en jongeren met ULRD functioneren vergelijkbaar met en zonder prothesen tijdens het uitvoeren van dagelijkse activiteiten. Door het gebrek aan functionele winst, het feit dat de prothesen te zwaar, moeilijk te gebruiken, of ongemakkelijk zijn, gebruiken 35-45% van de kinderen en jongeren met ULRD de prothese niet. De redenen waarom kinderen en jongeren met ULRD wel of geen prothese kiezen, zijn onvoldoende onderzocht. Ook is er geen feedback van deze kinderen en jongeren zelf beschikbaar over hun revalidatiezorg. Over alternatieve oplossingen, bedoeld om bepaalde activiteiten makkelijker uit te kunnen voeren, zoals adaptaties, is weinig literatuur beschikbaar. Tot nu toe is er geen onderzoek gedaan naar gebruik van adaptaties, hoe tevreden de kinderen en jongeren zelf zijn over adaptaties en of ze zich sociaal aangepast voelen met een adaptatie. De vragen die beantwoord moeten worden zijn: “Zijn kinderen en jongeren meer tevreden over prothesen of over adaptaties?” en “Is er een verschil tussen het sociaal aangepast voelen met een prothese in vergelijking met een adaptatie?”

Om betere keuzes te maken tijdens de revalidatie van kinderen en jongeren met ULRD, zouden behandelaars de werkelijke prestatie met de aangedane arm moeten testen. De Southampton Hand Assessment Procedure (SHAP) is een functionele test met een aantal voordelen ten opzichte van andere meetinstrumenten, zoals de universele toepasbaarheid bij de evaluatie van zowel (niet)aangedane als prothesehanden. Ook worden scores voor specifieke handgrepen onderscheiden. Hierdoor kan tijdens de revalidatiebehandeling gefocust worden op verbetering van handgrepen die met moeite uitgevoerd worden. Tenslotte kan met de SHAP snel en gemakkelijk informatie worden verzameld, want de test kan in 20 minuten worden afgenomen. De SHAP bestaat uit 26 taken, waarin gebruik gemaakt wordt van verschillende objecten. De psychometrische eigenschappen van de SHAP, moeten nog getest worden voor prothesegebruikers. Een SHAP voor kinderen en jongeren bestaat tot nu toe niet. Om SHAP bij kinderen te gebruiken, moeten de objecten en het protocol worden aangepast.

Dit proefschrift bestaat uit twee delen. Het doel van het eerste deel is om epidemiologische gegevens over kinderen met aangeboren afwijkingen van armen en benen (over het gehele spectrum van afwijkingen) in kaart te brengen, en om de effectiviteit en impact van prothesen en adaptaties bij kinderen en jongeren met ULRD te evalueren. In het tweede deel worden psychometrische eigenschappen van SHAP bij prothesegebruikers beoordeeld en daarna worden de ontwikkeling en het testen van de psychometrische eigenschappen van de aangepaste SHAP versie voor kinderen (SHAP children, SHAP-C) beschreven.

Het uiteindelijke doel van dit proefschrift was om uitgebreide kennis over het functioneren en meten van kinderen en jongeren met ULRD te verstrekken, waar zowel kinderen en jongeren met ULRD, onderzoekers als behandelaars baat bij hebben.

De International Classification of Functioning, Disability, and Health (ICF) werd gebruikt om het onderzoek in dit proefschrift te structureren. De ICF is een model dat door de World Health Organization wordt aanbevolen om gezondheid en beperkingen in functioneren te beschrijven. Dit proefschrift draagt bij aan de kennis over ICF componenten met betrekking tot kinderen en jongeren met ULRD, waaronder lichaamsfuncties en -structuren, activiteiten en participatie, persoonlijke- en omgevingsfactoren.

I. Kinderen en jongeren met ULRD–epidemiologie, functioneren, prothesen en adaptaties

Een gedetailleerde beschrijving van de Nederlandse bevolking met aangeboren afwijkingen aan de ledematen wordt in *hoofdstuk 2* als inleiding gegeven. De geboorte prevalentie van kinderen met een aangeboren afwijking aan de ledematen in Nederland blijkt 21,1 per 10.000 geboorten te zijn, gemeten over de periode 1981-2010. Hiervan zijn 6.9 per 10,000 geboorten reductiedefecten en bij 5 op de 10,000 geboorten is er sprake van ULRD. Deze ULRD waren meestal geïsoleerde defecten (geen deel van een syndroom of een specifieke afwijking).

In de *hoofdstukken 3 en 4* wordt kwalitatieve en kwantitatieve informatie over ICF componenten gegeven, waaronder: lichaamsfuncties, activiteiten, persoonlijke- en omgevingsfactoren. Kinderen en jongeren met ULRD rapporteerden dat zij moeite hebben met het uitvoeren van dagelijkse activiteiten, waarbij zij bij voorkeur hun aangedane arm, hun niet aangedane arm en andere lichaamsdelen gebruikten. Wanneer zij moeite hadden met

het uitvoeren van specifieke activiteiten, kozen ze vaker voor adaptaties dan voor prothesen om hun functionaliteit te verbeteren. De kinderen en jongeren gebruikten adaptaties om zelfzorg (hanteren van bestek), mobiliteit (aanpassingen aan fietsen), recreatie (sportactiviteiten) en hobby's (bespelen van een muziekinstrument) te faciliteren.

In online focusgroep interviews hebben kinderen en jongeren met een korte onderarm (waarbij de hand en een deel van de onderarm ontbreken), hun ouders en behandelaars hun mening gedeeld over de redenen om wel of niet prothesen te gebruiken en over de revalidatiezorg (*hoofdstuk 3*). De belangrijkste reden om een prothese te gebruiken was vanwege het cosmetische aspect. Een prothese hielp om het staren en ongewenste reacties van vreemden te verminderen en hielp bij de sociale integratie. Kinderen en jongeren die geen prothese gebruikten, vonden de prothese er niet mooi of onnatuurlijk uit zien of ze hadden de korte arm geaccepteerd. De prothesen werden beschouwd als nuttige accessoires voor specifieke activiteiten, zoals het tillen van zware voorwerpen of deelname aan sport. Toch konden veel van de kinderen en jongeren met ULRD de meeste activiteiten zonder prothese uitvoeren. Vaak genoemde redenen om geen prothese te dragen waren daarnaast extra gewicht, technische beperkingen en moeite met het bedienen van de prothese.

De rol van omgevingsfactoren, waaronder ouders, bij het gebruik van een prothese wordt in *hoofdstuk 3* toegelicht. Sommige ouders wilden hun kind graag "completer" zien en kozen daarom voor een prothese. Anderen waren van mening dat een prothese het functioneren van hun kind niet zou verbeteren en kozen daarom niet voor een prothese. De tevredenheid met adaptaties was significant hoger dan de tevredenheid met prothesen (*hoofdstuk 4*). Kinderen en jongeren met ULRD waren meer tevreden over de kenmerken van adaptaties, waaronder het geringe gewicht en het gebruiksgemak.

Revalidatiezorg lijkt aan de verwachtingen van kinderen en jongeren met ULRD en hun ouders te voldoen. Kinderen en jongeren met ULRD en hun ouders uitten hun ervaring met revalidatiezorg voor zover van toepassing (*hoofdstuk 3*) en waren tevreden over de revalidatiezorg, ongeacht de gebruikte oplossing (prothesen of adaptaties, *hoofdstuk 4*). Patiëntgerichte zorg wordt als een gewenste benadering in de revalidatie voorgesteld en contact met lotgenoten bleek belangrijk te zijn voor het uitwisselen van informatie, emotionele steun, en het omgaan met ULRD.

II. ULRD–functionele testen: SHAP en SHAP-C

Hoofdstuk 5 en 6 geven informatie over de psychometrische eigenschappen van SHAP, een functionele test waarmee de lichaamsfuncties van de aangedane arm worden geëvalueerd tijdens het uitvoeren van activiteiten van het dagelijks leven. De taken van SHAP werden beïnvloed door leereffecten, wanneer SHAP herhaaldelijk werd toegepast bij beginnende prothesegebruikers (dit waren niet aangedane proefpersonen die een prothese simulator gebruikten, *hoofdstuk 5*). Prestaties werden beïnvloed door warming-up: in een set van twee sessies duurde de eerste sessie langer dan de tweede. Dit effect wordt warming-up decrement genoemd, wat wil zeggen dat de prestaties in de eerste sessie minder goed zijn. Beginnende prothesegebruikers overschreden meerdere keren de tijdslimieten die voor elke taak opgelegd waren; dit had gevolgen voor zowel de functionele scores van de handgrepen als voor de totale score (Index of Function). Behandelaars moeten rekening houden met deze factoren, omdat die de prothese prestaties van beginnende gebruikers beïnvloeden.

De eerste stappen om SHAP voor kinderen (SHAP-C) aan te passen worden gepresenteerd in *hoofdstuk 6*. De verkleinde objecten bleken geschikt voor een kinderhand en ook voor een prothesehand. Alle proefpersonen (5 en 6-jarige kinderen zonder hand- of armafwijkingen) waren in staat om de SHAP-C taken uit te voeren. Deze taken kwamen overeen met de taken van de volwassen versie van SHAP. De intra-beoordelaar betrouwbaarheid, geëvalueerd met coëfficiënten van herhaalde metingen (repeatability coefficients), was relatief goed in vergelijking met de inter-beoordelaarsbetrouwbaarheid. In verdere studies over SHAP-C moet men rekening houden met mogelijke beïnvloedende factoren, zoals natuurlijke variabiliteit in prestaties van kinderen, wisselende motivatie, en reactietijden van de beoordelaars.

In *hoofdstuk 7* wordt een nieuw en eenvoudig scoresysteem voorgesteld om SHAP scores te berekenen (totaalscore en scores voor specifieke grepen). In het nieuwe scoresysteem, hebben de afzonderlijke taken een andere verhouding tot de normwaarden. In de originele SHAP worden de tijden van de taken gestandaardiseerd met behulp van z-scores en variabiliteit (standaarddeviaties) van de normwaarden die afgeleid zijn van SHAP-scores van niet aangedane proefpersonen. De standaarddeviaties van prothesegebruikers kunnen echter verschillen van die van niet aangedane mensen. Daarom stelden wij voor de SHAP scores anders te berekenen. In de nieuwe berekening is de standaardisatie van de tijden, die gemeten voor de verschillende SHAP taken zijn, onafhankelijk van de variabiliteit in de

normwaarden van niet aangedane proefpersonen. In toekomstig onderzoek zou moeten worden bepaald hoe de invloed van taken in de functionele score kan worden gewogen. Dit betekent dat de taken / handgrepen die vaker voor activiteiten van het dagelijks leven worden gebruikt, een grotere bijdrage aan de functionele scores zullen leveren. In SHAP leveren alle taken een vaste bijdrage aan de functionele scores en het overschrijden van de tijdlimiet van een bepaalde taak kan de functionele scores in grote mate beïnvloeden.

In het laatste hoofdstuk, *hoofdstuk 8*, is een algemene discussie opgenomen over de toepasbaarheid van de belangrijkste resultaten voor de praktijk binnen de gezondheidszorg en toekomstig onderzoek. Het gebruik van prothesen en adaptaties heeft impact op het fysieke en psychosociale functioneren van kinderen en jongeren met ULRD; ze voelen zichzelf meer als andere kinderen. De cosmesis en zelfstandigheid moeten worden benoemd door klinici die met kinderen en jongeren met ULRD werken. Dit omdat de cosmesis en zelfstandigheid helpen bij sociale integratie. Clinici en onderzoekers wordt geadviseerd kennis te nemen van de psychometrische problemen met SHAP, namelijk de leereffecten, en met SHAP-C, zoals de slechte betrouwbaarheid tussen beoordeelaars. Toekomstig onderzoek zou zich moeten richten op het verder ontwikkelen en verbeteren van deze instrumenten.

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Extremities, Pain and Disability

Missie: EXPAND draagt bij aan participatie en kwaliteit van leven van mensen met aandoeningen en amputaties van de extremiteiten of met pijn aan het bewegingsapparaat.

EXPAND omvat twee speerpunten: onderzoek naar aandoeningen aan en amputaties van extremiteiten met nadruk op stoornissen, activiteiten en participatie en onderzoek naar chronische pijn en arbeidsparticipatie. EXPAND draagt bij aan het UMCG-brede thema Healthy Ageing.

Research Department of Rehabilitation Medicine

Center for Rehabilitation UMCG

EXPAND

Extremities, Pain and Disability

Mission: EXPAND contributes to participation and quality of life of people with conditions and amputations of the extremities and musculoskeletal pain.

EXPAND focuses on two spearheads: research on the conditions and amputations of the extremities with emphasis on body functions and structures, activities and participations, and chronic pain and work participation. EXPAND contributes to Healthy Aging, the focus of the UMCG.

