Consequences of success in pediatrics: young adults with disability benefits as a result of chronic conditions since childhood

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General introduction and outline of the thesis
This thesis is about the psychosocial functioning, the emotional well-being and the factors affecting the vocational success and well-being of young adults with disability benefits as a result of a chronic somatic condition since childhood. The following paragraphs explain the process of growing up with a chronic somatic condition and the additional challenges it entails. The added value of work participation in general, the work situation of people with chronic conditions and an explanation of the disability benefits are described below. This introduction continues with the research questions, the research model and gives an elaboration of the research topics.

CONSEQUENCES OF SUCCESS IN PEDIATRICS

In the past, most childhood diseases were fatal for the majority of children. As a result of improved medical knowledge, technology and practices, including the development of pediatric surgical interventions, pediatric cardiac surgery and pediatric anaesthesiology, pediatricians were able to change the natural course of numerous diseases. This resulted on the one hand in higher survival rates for many diseases, including cancer, metabolic diseases, congenital heart disease and cystic fibrosis 1,2, but on the other, in an increased number of children growing up with chronic health problems and morbidities since these children have been treated but have not been cured. In addition, the prevalence of some diseases like obesity, asthma and attention deficit hyperactivity disorder has increased as well 1.

Consequently, the number of children growing up with a chronic illness has increased enormously in recent decades. Although numbers vary depending on methods and definitions, by any estimate the scope of the problem is huge. The number of chronically ill children approximates 7 to 20 percent of all children 1,3 and will likely further increase because of medical advancements 1. In the Netherlands there are about 500,000 children (14%) with a chronic disorder 1. The prevalence of specific disease and conditions range widely. In the Netherlands, the most common diagnosed illnesses in childhood are asthma (10%) and eczema (6%). Other moderately prevalent illnesses (<1%) are diabetes mellitus type 1, epilepsy, juvenile idiopathic arthritis (JIA), sickle cell anemia, cystic fibrosis, hemophilia, and inflammatory bowel diseases (CBS statistics Netherlands, 2013).

With the increased rates of successful treatment and its positive effect on life expectancy, new questions emerge. Now survival is no longer the major challenge, additional challenges are added. The focus has shifted to the influence of growing up with a chronic condition and to how pediatric illness affects adult life. What happened to those with a childhood onset illness or disability while growing up? And how do they function in adult life? Are they independent and
successful? Are they happy? What help do they need? They live longer, they mature, but what does it mean to grow up with a chronic illness for later societal participation as an adult in our society?

GROWING UP
Growing up and learning to be an adult is for everyone a complicated process wherein people during childhood, adolescence and young adulthood experience multiple transitions\(^4,5\). These transitions are processes during which youth develop attitudes, behaviors, and skills to successfully move to adulthood\(^6\). The transitions to adulthood comprise different developmental tasks, e.g. the consolidation of their identity, the transition from family life to independent living, and from education to employment. Some transitions are determined by age or development, while others are dictated by ‘systems’, for example, the educational system or healthcare system\(^5\). This journey from childhood to adulthood is characterized by starts and stops, anxiety and anger, and hope and despair, coupled with an ever-increasing mastery of an essential set of life skills necessary for independence in adulthood\(^7\). Youth development is much more complex than biological growth and maturation. There are patterns of physical, cognitive, social and emotional changes that youth go through, but these changes often occur simultaneously or in rapid succession\(^5\). In the 21st century, youth development and transition into adulthood have become less straightforward\(^5,8\).

Arnett\(^9\) has described a new phase of development called ‘emerging adulthood’, a period that exists only in highly industrialized or post-industrial cultures that postpone the entry into adult roles and responsibilities until well past the late teens. Such countries require a high level of education and training for entry into the information-based professions that are the most prestigious and lucrative, so many of their young people remain in school into their mid-20s. Marriage and parenthood are typically postponed until well after schooling is completed, permitting a period of exploration of various relationships before marriage and for exploration of various jobs before taking on the responsibility of supporting a family. These young people between the ages of 18 and 30 are on the threshold of adulthood\(^6\). Emerging adulthood is also notable for its heterogeneity. It is the time when people’s lives are least likely to be structured by social institutions\(^10\). Children’s and adolescents’ lives are structured by family and school, and adults lives (in young adulthood and thereafter) by family and work. However, emerging adults typically move away from their families, and their school and work patterns tend to be formed, but are relatively unstructured and unstable\(^9\). The characteristics of this developmental stage include the following: identity explorations – trying out various possibilities in love, work, li-
ving situations and world views; instability; being self-focused and feeling in-between—neither adolescent nor adult; and possibilities—when hopes flourish and young people have unparalleled opportunities to transform their lives. These are not features that exist only in emerging adulthood, but they are more pronounced in emerging adulthood than at other ages. 

GROWING UP INTO ADULTHOOD WITH A CHRONIC DISEASE OR DISABILITY (1)

Children and adolescents with chronic conditions are expected to pass through similar developmental stages as their nondisabled peers, to leave home, develop psychosocially, and define a role for themselves in the community through employment or other activities. A chronic condition can have a profound influence on aspects of development during the transition into adulthood. Children and adolescents with chronic conditions face many additional challenges during this process. Most children experience several consequences of their health care needs; they depend on medical care or medical aids to some extent, varying from medication use to medical interventions or medical aids (e.g., daily inhalation of corticosteroids, intravenous alimentation, and wheelchair). Some of them also have to deal with late treatment effects. Further, transition into adulthood may be more difficult as there will be less exposure to everyday events and therefore to opportunities to learn and explore. Social opportunity is lessened by ill health including frequent hospitalizations, and exclusion from the opportunities afforded by mainstream schooling or unsupervised play with in the age-appropriate peer group. The adolescent process of exploring limits, reality testing, and self-image development may be severely delayed. Enhanced vulnerability owing to physical or psychological stress may also complicate the transition to adulthood.

Concern has risen about the implications of growing up with a chronic condition. The condition and late treatment effects may affect the physical functioning and emotional functioning. Children with chronic diseases are somewhat more likely than healthy children to show maladjustment. They tend to suffer more than average from behavior problems, especially internalizing problems such as depression, anxiety, and social withdrawal. A meta-analysis of Pinquart and Teubert found children and adolescents with chronic physical illness to have, on average, lower levels of academic, physical, and social functioning than their healthy peers. Impairments were largest for physical function-
ing and smallest for social functioning. In line with this, evidence suggests that school-aged children with chronic conditions, regardless of their diagnosis, are at risk for lower participation in everyday activities than their peers; they participate less frequently in almost all activities compared with children without a physical disability. Furthermore, it is known that youth who have grown up with a disease are at risk for a delay in age-specific developmental tasks compared with peers, resulting in a delayed course of life. In summary, a part of the children with chronic conditions may have difficulties while growing up.

Except for survivors of childhood cancer and those with end-stage renal disease, little research exists about whether the challenges and gaps between youth with chronic conditions and those without persist into adulthood. Will they finish education, get jobs, and live independently? Can they become autonomous in a society where self-sufficiency and independence receive widespread recognition and social endorsement? And most important, how do they fulfill their lives in a meaningful and satisfying way? Given that the majority of research on emerging adulthood focuses on typically developing, college-attending healthy youth, little is known about the impact of growing up with a chronic condition on the milestones of emerging adulthood including work participation and the adaption to adult life.

**WORK**

An important way to participate in social life is work. Work generates material and immaterial benefits. The material purpose of work is earning an income to be able to support oneself. Besides money, employment offers a lot of other additional immaterial benefits like the possibility for further self-development, social relationships, development of skills, daily rhythms, and often, self-esteem and meaning in life.

Having a paying job at some time during high school has become a near-universal adolescent experience. At first, paid work is episodic and generally quite limited. Many youth start to work informally even earlier, at about the age of twelve, most often in their own neighbourhoods, babysitting, cutting grass, or doing various odd jobs. By the age of 16, adolescent workers are more likely to have formal jobs, working in the retail and service sectors of the economy, for example in fast food restaurants, or retail stores. Employment becomes more regular and more time-consuming during the latter years of high school, with many teens working 20 or more hours per week. During and after their study this process continues; most young adults start a working career which continues in adulthood.
Somatic conditions and work participation

Having a chronic disease or disability often negatively affects the capacity to participate in gainful employment. A poor health condition, however, does not necessarily imply exclusion from the labour market. Yet, in various studies, the labour market position of people with a chronic disease or disability is found to be problematic, with regard to getting a job as well as job continuation. This, in turn, leads to a variety of economic, social and quality of life problems. The consequences of work disability for patients with chronic diseases include financial difficulties, increased social isolation, decreased confidence and self-esteem and stress. Besides, several studies among people with chronic somatic diseases show lower Health Related Quality of Life (HRQoL), worse mental health and more anxiety and depression in non-workers than in those who are participating in the workforce.

Adolescents and young adults with somatic conditions and work participation

Most of the research on the work experiences of people with disabilities has focused on adults while much less attention has been paid to younger people just entering the workforce. However, employment and its skill-building opportunities are essential elements for a successful transition to adulthood for young people. Adolescents want to work to earn their own spending money to be able to buy the accoutrements of adolescent life and take part in the leisure activities among their peers. Working enriches young people’s life experiences, thereby enhancing self-confidence, self-discipline, self-esteem, and independence. Furthermore, working experiences help young people think about their possible career trajectories and develop work attitudes. The shift out of school into education and occupation is a developmental stage in which young people gain increasing sense of control over their lives, experience a greater responsibility, and form an identity which they can use to shape their sense of self that will likely endure throughout adulthood. Avoiding unemployment in early adulthood is particularly important, because the decisions and actions that occur during this time, can affect income levels and occupational achievement across the lifespan.

In general, youth with disabilities are much less likely to be employed than their peers during high school and often encounter barriers when striving for independence. This trend of unemployment during adolescence appears to be continuing in young adulthood. Young adults with disabilities experience considerably lower employment rates compared to their non-disabled peers and their unemployment periods often last longer and they face higher risks of losing their jobs than non-disabled people.
YOUNG ADULTS WITH SOMATIC CONDITIONS AND DISABILITY BENEFITS IN THE NETHERLANDS

In the second half of the 1970s the Netherlands government made a crucial decision to support that young people with chronic conditions or disabilities can participate in society with a personal income, independent from parents and others. This was effected through the General Disability Act (AAW), a national insurance against disability, which in principle all residents of Netherlands were insured for. There have been a number of developments and changes in this insurance for young people over time. The following Act was effective during this study:

The Disablement Assistance (Young Persons) Act; ‘old’ Wajong (2)

The General Disablement Act was abolished in 1997, and on 1 January 1998 a new Act came into force for young people and students who have become disabled at an early age and therefore do not qualify for wage-related benefits: the Disablement Assistance (Young Persons) Act, known as ‘Wajong’ in Dutch. This Act provides (supplementary) income support as well as support to find employment and if necessary support at the work place. The benefit is not obligatory or automatically including all young adults with disabilities: the initiative for application lies with the individual and his/her social environment. A Wajong benefit is payable not earlier than the 18th birthday, for as long as the inability to work lasts and ends when the recipient reaches the age of 65. The Employee Insurance Agency (UWV) implements the Act and is also responsible for determining the level of benefit paid. The level of benefit received under the terms of the Act depends on age and the amount someone can earn from a job; Wajong income support is a supplementary payment on top of what a young person with a chronic disease or handicap is able to earn from work. The UWV insurance physicians assess the work ability of the claimant considering the medical history and prognosis and describe the limitations for work. Based on the limitations assessed by the insurance physician, employment specialists investigate what jobs the claimant could perform on the labour market despite these limitations and what work opportunities are available. Depending upon the question whether or not the young disabled can theoretically earn a minimum wage with a job, a disability pension will be refused or awarded. A part of those who receive a Wajong benefit is potentially (partially) capable to work. The other part is declared fully unfit for work.

(2) The new Wajong Act was introduced on 1 January 2010. See appendix for further details.
**Numbers of Wajong applicants**

The numbers claiming benefits under the Wajong scheme rose from 40,000 in 1976 to 127,400 in 2000 and 238,700 in 2013 (table 1).\(^{50,51}\) This meant that one out of every 20 people aged 18 (4.8%) was claiming and receiving this disability benefit.\(^{52}\) This increase contrasted with the numbers of claims made under the WAO/WIA Act – which caters for people who develop a disability at some stage during adulthood – which has seen modest falls in recent years. A number of studies have examined the possible causes of this take-up of the scheme. The reasons behind the increase in claimants of this disability benefit are complex and likely to be multi-factorial. For example, the eligibility rules for entry into benefits systems are likely to have changed, diagnostic criteria may also have changed, and systems may have become more efficient at recognising health problems. At a personal level, there may have been a real increase in health problems or there may have been a decline in employment prospects which has pushed people to register as disabled. Social reasons, especially those relating to the level of family support, may also play a role. Also, statistical or administrative aspects, the maturation of the scheme, and greater awareness of the scheme amongst potential claimants can have influence. In addition, the current economic climate could affect attitudes towards the employment of young people with health problems. It is not known what proportion of children and young people who grow up with a chronic somatic disease or disability eventually ends up in the Wajong.

**TABLE 1: Number of recipients of Wajong benefits (2003 – 2013)**

<table>
<thead>
<tr>
<th></th>
<th>Influx Wajong benefit</th>
<th>Outflux Wajong benefit</th>
<th>Current benefits end of year</th>
</tr>
</thead>
<tbody>
<tr>
<td>2003</td>
<td>8.200</td>
<td>4.400</td>
<td>138.500</td>
</tr>
<tr>
<td>2004</td>
<td>9.400</td>
<td>4.900</td>
<td>142.600</td>
</tr>
<tr>
<td>2005</td>
<td>10.400</td>
<td>5.600</td>
<td>147.200</td>
</tr>
<tr>
<td>2006</td>
<td>13.600</td>
<td>4.900</td>
<td>155.900</td>
</tr>
<tr>
<td>2007</td>
<td>15.300</td>
<td>4.400</td>
<td>166.800</td>
</tr>
<tr>
<td>2008</td>
<td>16.100</td>
<td>4.300</td>
<td>178.600</td>
</tr>
<tr>
<td>2009</td>
<td>17.600</td>
<td>4.300</td>
<td>192.000</td>
</tr>
<tr>
<td>2010</td>
<td>17.800</td>
<td>4.600</td>
<td>205.100</td>
</tr>
<tr>
<td>2011</td>
<td>16.300</td>
<td>5.200</td>
<td>216.200</td>
</tr>
<tr>
<td>2012</td>
<td>15.300</td>
<td>5.100</td>
<td>226.500</td>
</tr>
<tr>
<td>2013</td>
<td>17.700</td>
<td>5.500</td>
<td>238.700</td>
</tr>
</tbody>
</table>

Annual report Employee Insurance Agency (UWV jaarverslag 2013)
Gender, age and diagnoses of applicants at Wajong influx

Table 2 shows the gender, age and diagnoses of new Wajong applicants. There are slightly more men than women applying for a Wajong benefit. Most applicants enter the Wajong at an early age. The Wajong influx contains for the most part of youngsters having developmental disorders. Another large group of young people have been diagnosed with psychiatric disorders (25%, for instance schizophrenia or personality disorder). These statistics only partly reflect the reality because 42% have multiple conditions diagnosed. About 14 percent is having a disability benefit because of somatic (physical) malfunctions or disorders.

<table>
<thead>
<tr>
<th>Diagnosis group</th>
<th>2008</th>
<th>2010</th>
<th>2013</th>
</tr>
</thead>
<tbody>
<tr>
<td>Development disorders</td>
<td>60</td>
<td>66</td>
<td>67</td>
</tr>
<tr>
<td>Mental retardation (intellectual impairments, learning problems)</td>
<td>38</td>
<td>37</td>
<td>35</td>
</tr>
<tr>
<td>Autistic spectrum impairments</td>
<td>11</td>
<td>15</td>
<td>18</td>
</tr>
<tr>
<td>Attention deficit (ADHD, ADD)</td>
<td>4</td>
<td>8</td>
<td>8</td>
</tr>
<tr>
<td>Other development disorders</td>
<td>6</td>
<td>5</td>
<td>5</td>
</tr>
<tr>
<td>Psychiatric diseases</td>
<td>26</td>
<td>21</td>
<td>19</td>
</tr>
<tr>
<td>Somatic diseases</td>
<td>15</td>
<td>13</td>
<td>14</td>
</tr>
</tbody>
</table>

Employee Insurance Agency Wajong monitor 2008 en 2010; key data Wajong 2013

Work participation of Wajong applicants

Although some of the young adults with Wajong benefits were unable to work in any way because of their limitations, others could and were willing to work. However, those young adults with a Wajong benefit who were able to work remain under-represented in the labour market. Data from the Social and Economic Council of the Netherlands (SER) indicated that around 60% of people on Wajong benefit were able to work (at least 12 hours a week). In reality, around 25% did so (table 3). The greatest part of the young adults with Wajong benefits has difficulty participating in employment. The problem of the low em-
ployment rate among people on Wajong benefit is in finding employment, as well as in holding on to it. A high proportion of those who find employment lose their jobs within a short time. Around 20% of the people who are not working when they get the Wajong disability benefit find a job within a year. Almost half of those who find a job, lose their job again within one year.

**TABLE 3: Number of working people with a Wajong benefit**

<table>
<thead>
<tr>
<th>Year</th>
<th>Number of people with a Wajong benefit</th>
<th>Number of working people with a Wajong benefit</th>
<th>Percentage working people with a Wajong benefit</th>
</tr>
</thead>
<tbody>
<tr>
<td>2005</td>
<td>147,161</td>
<td>38,733</td>
<td>26.3%</td>
</tr>
<tr>
<td>2006</td>
<td>155,860</td>
<td>39,814</td>
<td>25.5%</td>
</tr>
<tr>
<td>2007</td>
<td>166,805</td>
<td>45,613</td>
<td>27.3%</td>
</tr>
<tr>
<td>2008</td>
<td>178,590</td>
<td>45,900</td>
<td>25.7%</td>
</tr>
<tr>
<td>2009</td>
<td>191,956</td>
<td>47,598</td>
<td>24.8%</td>
</tr>
<tr>
<td>2010</td>
<td>205,134</td>
<td>49,700</td>
<td>24.2%</td>
</tr>
<tr>
<td>2011</td>
<td>216,240</td>
<td>53,844</td>
<td>24.9%</td>
</tr>
<tr>
<td>2012</td>
<td>226,481</td>
<td>53,749</td>
<td>23.7%</td>
</tr>
<tr>
<td>2013</td>
<td>238,708</td>
<td>53,890</td>
<td>22.6%</td>
</tr>
</tbody>
</table>

*Annual report Employee Insurance Agency (UWV jaarverslag 2013)*

**THE EMWAJONG STUDY**

Among the young adults with Wajong benefits as a result of a chronic illness or disability, some have a congenital disease or disability, some are survivors of a severe disease in childhood, some have a long-standing yet less severe condition and some have been ill or disabled since adolescence only. Health status and functional limitations, however, do not tell us everything about the impact of growing up with a chronic condition on well-being and participation in society. The major question is that of their well-being, independence and future. Since the Wajong population can be considered as the most vulnerable young adults with chronic conditions – those who have to apply for disability benefits as a result of their conditions – it is essential to gain insight into the challenges they may be encountering in dealing with their condition while becoming an adult. All in all, the (often hidden) psychosocial consequences of the pediatric success story on growing up and social integration deserves attention. If we can gain insight into the adjustment and process of growing up of the Wajong population, their emotional well-being and factors affecting their vocational careers, we may be able to develop strategies to support this vulnerable population towards adulthood independence and economic participation. It is crucial that these young adults in our society are able to find their place in the adult world.
Aim and research questions

The purpose of this study was to evaluate the psychosocial functioning, the emotional well-being and the factors affecting the vocational success and well-being in young adults with a Wajong benefit for a childhood-onset chronic somatic condition while growing up. The study was aimed at young adults with a disability benefit as a result of a somatic illness or condition. The primary research questions of this study are as follows:

1. How does the psychosocial developmental trajectory (course of life) of young adults with disability benefits as a result of a somatic condition compare to that of a reference group?
2. How does the Health Related Quality of Life (HRQoL) and anxiety and depression of young adults with disability benefits as a result of a somatic condition compare to that of reference groups?
3. What are the Illness Cognitions (IC) of young adults with disability benefits as a result of a somatic condition? Is there an association of IC with HRQoL, anxiety and depression in this study population?
4. Is there a difference in the psychosocial development trajectory of young adults with and without a disability benefit?
5. What are the disease characteristics, work characteristics, and work motivation of young adults with disability benefits as a result of chronic conditions and the association of those factors and IC with the likelihood of being employed?

Research model

The guiding framework (figure 1) for this thesis is the International Classification of Function, Disability and Health (ICF) (World Health Organisation, 2001). This model integrates the major models of disability - the medical model and the social model - as a bio-psycho-social synthesis and measures health and disability at both individual and population level and covers the entire life span. The ICF classification does not classify people according to a diagnosis but describes different ways of functioning. The model identifies three levels of human function: body functions and structures (perspective of the body), activities (perspective of the individual), and participation (perspective of society). The functioning (both the capacity and the performance) of the individual can be influenced by the disease or disorder, but also by personal factors (e.g. age, gender, self-esteem and motivation) and by environmental factors (e.g. the social environment). ‘Disability’ is the ICF umbrella term for impairments (a problem in bodily functioning), activity limitations (like walking or personal care) and participation restrictions (a restriction an individual may experience in involvement.
in life situations, like labour, going to museums, going on holidays and meeting friends) 55, 56, influenced by personal and environmental factors.

**FIGURE 1: The international Classification of Functioning, Disability and Health (ICF)**

The ICF model conceptualises functioning and disability as a dynamic interaction; disability can lead to loss in abilities, which in turn can lead to problems in participation in society. Young adults with disability benefits because of chronic conditions may experience problems in everyday life. They may have participation restrictions while growing up because of difficulties in performing desired activities, which can influence their functioning and health condition. Outlined below are the topics in this study that could influence the interrelation between the functioning and disability of young adults with Wajong benefits.

**Medical parameters and socio-demographic factors**

The influence of socio-demographic factors on adjustment is often studied in pediatric literature. For instance, boys tend to have more externalizing problems than girls 57. Furthermore, higher socio-economic status, higher educational level of the parents, higher income, and belonging to a non-minority were protective factors associated with child and parent adjustment 58. A number of illness severity parameters are found to be associated with psychosocial problems, such as: the intensity of the treatment and hospitalization 59, visible symptoms of illness 60, 61, limitations in school attendance 60, and a poor or uncertain disease prognosis 62. However, despite these empirical associations with illness characteristics, most studies that include both illness- and psychosocial risk-factors in a multivariate regression model show that illness factors account...
for little of the variability in psychosocial adjustment compared to psychosocial factors. Less is known about medical and socio-demographic parameters and adjustment in emerging adulthood. A meta-analysis of Pinquart indicated that stronger differences in the achievement of developmental milestones - finishing advanced education, finding employment, leaving the parental home, marrying, and becoming parents - were found for respondents with neurological illnesses and sensory impairment than in individuals with other chronic diseases. Lower success rates were also observed if the illness/disability is highly visible to others and in the case of longer illness duration.

**Course of Life**

The developmental consequences of growing up with a childhood chronic condition may have consequences in adulthood. The fulfilling of age-specific developmental tasks in youth is of great importance to the adjustment in adult life. The developmental tasks and the resulting developmental milestones that are necessary in the development of a child are referred to as ‘the course of life’ (CoL). The normal developmental tasks of childhood and adolescence involve the attainment of social and academic competence, the development of peer relationships and increasing independence from parents. Children and adolescents with chronic diseases are expected to pass similar developmental stages as their nondisabled peers. For patients with impairments, reaching these developmental stages can be challenging. They face many additional challenges to negotiate and obtain normative developmental milestones and maintain adaptive functioning. The burden of the disease or disability, treatment, hospitalization and long-term medical sequelae, and decrease in social and school-based participation could be a threat to the accomplishment of developmental tasks, resulting in a delayed course of life.

From a developmental psychological point of view, risk behaviour is also relevant. To a certain extent, displaying risk behaviour - in terms of trying out - is part of the development from being a teenager to becoming an adult. Adolescents grown up with a chronic condition may display less risk behaviour than do their healthy peers, because they are keenly aware of the vulnerability of their health. Moreover, increased parental involvement as a result of the paediatric condition experience may limit children’s opportunities to have unsupervised time with peers, which may decrease their opportunities to engage in risk activities with peers. On the other hand, we could possibly expect to observe more risk behaviour among adolescents with a childhood-onset, in compensation for the limitations that were imposed upon them by disease in their youth. The course of life has been investigated in several disease specific groups.
for a delayed CoL compared with the peer group. Particularly the CoL of patients with end-stage renal disease and survivors of childhood cancer was found to be hampered. So far, little is known about the CoL of young adults with somatic chronic diseases or disabilities with disability benefits.

Health Related Quality of Life

WHO defines Quality of Life (QoL) as an individual’s perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns. It is a broad ranging concept affected in a complex way by the person’s physical health, psychological state, personal beliefs, social relationships and their relationship to salient features of their environment (www.who.int). Health-related quality of life (HRQoL) refers to how health impacts on an individual’s ability to function and his or her perceived well-being in physical, psychological and social domains of life. Physical functioning refers to activities of daily living, as well as to physical symptoms resulting from disease or treatment. This physical functioning part of HRQoL includes basic activities, such as self-care (eg, bathing, dressing), as well as work-related activities (whether paid or not) such as housework and career. Social functioning refers to social relationships and interactions, and to societal integration. The psychological well-being part of HRQoL is somewhat more subjective than the functioning part, because it relies almost exclusively on the internal, subjective perceptions of the respondent. Psychological functioning ranges from severe psychological distress to a positive sense of well-being, and it may encompass cognitive functioning. Included is whether the person feels happy, sad, depressed, or anxious (emotional well-being), whether they are in severe pain or have no pain at all, and whether they are energetic or lethargic. Beyond this core set of HRQOL domains, additional issues may be relevant. In the case of young people with chronic illness, developmental changes should be taken into consideration. Definitions of HRQoL therefore should include goals and aspirations as well as the constraints imposed through ill-health and treatment.

The past 20 years has seen an explosion of studies of HRQoL in with chronic conditions, reflecting a widening of focus from biomedical outcomes to include psychological and social dimensions of health. This expansion in the field of HRQoL studies is mainly focussed on children and adults. Overall, children with a chronic disease are at a greater risk of HRQoL problems than their healthy peers, but not in all cases, and not on all domains. A lot of studies describe the HRQoL of adolescents with a chronic childhood condition, but less studies report the HRQoL of young adults with a chronic childhood condition. Most studies on HRQoL on young adults with a chronic condition have been
illness specific and show affected HRQoL scores \(^{81, 82}\). For example, survivors of childhood cancer reported a lower HRQoL than their peers \(^{81}\) and specific chronic health problems had a negative impact on survivors’ HRQoL \(^{83}\). The literature on adults with chronic illness since childhood found a lower HRQoL and more emotional problems compared to the general population \(^{12-15}\). Socio-demographic characteristics such as gender, age and education are generally of little significance for the general health perception of adults with a chronic illness or disability. The severity of the condition seems to have more impact on their HRQoL than the nature of the condition itself \(^{16}\).

Having a paid job can have positive effects on the wellbeing and QoL of people of working age. Several studies among people with chronic somatic diseases show lower Health Related Quality of Life in non-workers than in those who are participating in the workforce \(^{37-40}\). Nevertheless, there are also studies indicating only small differences in the HRQoL between working and non-working people with a chronic somatic disease \(^{56, 84}\). Particularly, the severity of the physical disability carries weight in the evaluation of someone’s HRQoL. Participation in work and well-being appears to be less associated when adjusted for the severity of physical disability \(^{56}\). However, few studies have focused on the emotional well-being of young adults with childhood onset chronic conditions who encounter barriers when pursuing employment, as compared to young adults without chronic conditions. Also, the HRQoL and emotional functioning of young adult beneficiaries with a childhood-onset somatic condition as a group has never been studied.

**Anxiety and Depression**

In the general adult population, having a chronic illness is a significant risk factor for the development of mental health disorders such as anxiety and depression \(^{85}\). Internalizing problems, such as anxiety and depression are also described as potential concerns for children with chronic illness \(^{86}\). Several meta-analyses on children with chronic illness have found elevated levels of anxiety and depression in children with chronic physical illness \(^{87, 88}\). Two meta-analyses of Pinquart and Shen \(^{89, 90}\) compared levels of anxiety and depression of children and adolescents suffering from chronic illnesses with their healthy peers across a large number of diseases. Strongest elevations were found for chronic fatigue syndrome, migraine/tension headache, sensory impairment and epilepsy in relation to anxiety, and in chronic fatigue syndrome, fibromyalgia, cleft lip and palate, migraine/tension headache, and epilepsy in the case of depression. Most research on mental health and chronic conditions concern children or adult populations. Less is known about anxiety and depression on those in the period of emerging adulthood.
Illness cognitions

There is increasing evidence that psychological factors play an important role in the adaptation to living with a chronic disease. Studies on adaptation to chronic illness have shown that there often is a considerable discrepancy between the level of illness-related dysfunction as reported by patients and the underlying pathology of their disease. The magnitude of the physical, mental, and social problems that patients with chronic diseases present can vary greatly from patient to patient, even in patients with the same medical condition or the same severity of disease. The weak relationship between biomedical parameters and wellbeing has given rise to the hypothesis that other, psychosocial, factors contribute to health outcomes in patients with chronic diseases.

In recent years, the role of illness perceptions and coping responses of patients have especially been highlighted. Health psychologists have shown that, to make sense and respond to the problems caused by a chronic illness, patients create their own explanations or ‘beliefs’ on their illness. When patients are diagnosed with an illness they generally develop an organised pattern of beliefs about their condition. These illness perceptions or cognitive representations directly influence the individual’s emotional response to the inherently aversive character of a chronic condition, to maintain a sense of balance and to achieve a satisfying quality of life. This in turn determines how patients respond to the chronic condition; in their coping behaviour such as adherence to treatment. It has been commonly assumed that the way adult patients perceive and think about their illness accounts for much of the individual differences in their physical and psychological health status. Specifically, patients who report high levels of helplessness and low levels of acceptance with regard to their illness, emphasize the negative aspects of their condition. They generalize their cognitions to all facets of daily life and consequently experience worse physical and psychological functioning. In this view, illness cognitions (IC) can be considered as prognostic factors predicting physical functioning, psychological distress and therefore possibly adaption to society.

In addition, there is increasing evidence that illness cognitions could play a role in work participation. A review study of Hoving et al. exploring the relationship between illness perceptions and work participation in patients with somatic diseases and complaints found promising evidence. Even though the number of studies in the review was limited, all included studies report significant findings between one or more illness perception dimensions and measures of work participation. In particular, all studies found that non-working people perceive more negative consequences of their disease. This suggests that illness perceptions play a role in the work participation of patients with somatic diseases or complaints. Most of the research on IC has focused on adults while
much less attention has been paid to younger people who grew up with a chronic condition. Also, the IC of young adult beneficiaries as a group has never been studied.

Factors that influence participation in work
There is little detailed information about the employment patterns of young people with chronic conditions compared to their non-disabled peers. Although there are studies on (return-to-)work factors among adults with chronic conditions, the factors found for impaired adults may not be the same as those for young adults who are at the beginning of their career. Knowledge regarding factors that influence the work participation of these young adults in practice is hardly available. Previous studies mentioned the following factors: socio-demographics (age, gender, education, geographic location and the number of people living in the household), psycho-social functioning, severity of disability, type and duration of disability, level of depression and dispositional optimism. A systematic review concerning factors that promote or hinder work participation in young adults with disability benefits found motor impairment, low physical ability, co-morbidity, epilepsy, IQ lower than 80, younger age at diagnosis and higher radiation grade in cancer survivors to be related to negative employment outcome. However, the abovementioned studies focussed on specific disease categories or examined both physical and mental conditions. So far, little is known about the early determinants of work integration of young adults with disability benefits because of a chronic somatic condition as a group.

Motivation
Young people’s motivation points out to be a critical ingredient in achievement in education and in occupational outcomes later in life. Those with a firmer work motivation are more likely to participate successfully on the labour market. Among the young adults with chronic conditions, motivation likely leads to greater initiative to search for employment. In addition, motivated people receive more guidance or support to find a job, and coaches and employers react positively to motivated individuals. However, lack of motivation has been found to be an internal barrier to employment for these young adults. It is hypothesised that motivation and early employment history are interrelated, such that early frequent job losses had an impact on the individual’s self-confidence and increased anxiety, which led to a likelihood of a pattern of job loss.

The work motivation of young adults with Wajong benefits as a result of somatic complaints has never been studied. What about their work motivation?
Are their aspirations associated with their illness cognitions and better employment outcomes in early adulthood?

**STUDY DESIGN**

The EMWAjong study is a large cross-sectional study. All young adults between 22 and 31 years of age who claimed a Wajong benefit (3) in the year 2003 or 2004 for a chronic somatic condition were invited to fill in an online questionnaire via a letter. The target group is a heterogeneous group with different types of health conditions and different abilities and limitations. Those with no sustainable work opportunities (classified as fully incapable for work) were excluded because the EMWAjong study aimed to identify factors that could help to improve vocational success. Those with serious cognitive impairment or psychiatric conditions were also excluded because the EMWAjong study was directed at young adults with childhood-onset somatic illness or disability.

In total, 2,046 persons were invited to take part in the study. To maintain the privacy of the beneficiaries, the invitation letter was sent by UWV, the Dutch benefits agency. The letter contained a personal log-in code, a password and a link to the online questionnaire. After two weeks, participants received a reminder letter. Participants who completed the entire questionnaire received a gift voucher. The sample was recruited at the end of 2009. The study was performed according to the regulations of the medical ethical committee; due to the once-only internet-based nature of the survey, no formal approval by the medical ethics committee was required.

The standardized and validated questionnaires that were used in the EMWAjong-study, related to the study questions, are presented in table 4. Due to privacy reasons, no information about the chronic conditions of the participants was provided by the benefits agency. This information was therefore derived through beneficiaries’ self reports. The questions concerning disease-characteristics were based on existing questionnaires and recommendations of experts in the field. Work participation was defined in conformity with Statistics Netherlands (CBS), as performance of paid work for at least 12 hours per week for an uninterrupted period of 6 months.

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(3) During this study, people who were not able to earn as much as a healthy person with the same qualifications and experience were classified as being occupationally disabled under the terms of Wajong. Those eligible for Wajong benefit were at least 25 per cent occupationally disabled on their 17th birthday or were younger than 30 and became 25 per cent occupationally disabled during the time as a student, as a result of which they were unable to work on a full-time basis after their studies.
**TABLE 4: The standardized and validated questionnaires used in the EMWAjong-study**

<table>
<thead>
<tr>
<th>Research question</th>
<th>Purpose</th>
<th>Measures</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 (Chapter 2 and 4)</td>
<td>How does the courses of life young adults with disability benefits as a result of a somatic condition compare to that of a reference group?</td>
<td>Course of life questionnaire (LVJV) 102</td>
</tr>
<tr>
<td>2 (Chapter 3 and 4)</td>
<td>How does the Health Related Quality of Life (HRQoL) and anxiety and depression of young adults with disability benefits as a result of a somatic condition compare to that of reference groups?</td>
<td>RAND-36 103, Hospital Anxiety and Depression Scale (HADS) 104</td>
</tr>
<tr>
<td>3 (Chapter 5)</td>
<td>What are the Illness Cognitions (IC) of young adults with disability benefits as a result of a somatic condition? And is there an association of IC with HRQoL, anxiety and depression in this study population?</td>
<td>Illness Cognition Questionnaire (ICQ) 92, RAND-36, Hospital Anxiety and Depression Scale (HADS)</td>
</tr>
<tr>
<td>4 (Chapter 6)</td>
<td>Is there a difference in the psychosocial development trajectory of young adults with and without a disability benefit?</td>
<td>Course of life questionnaire (LVJV)</td>
</tr>
<tr>
<td>5 (Chapter 7)</td>
<td>What are the disease characteristics, work characteristics, and work motivation of young adults with disability benefits as a result of chronic conditions and the association of those factors and IC with the likelihood of being employed?</td>
<td>Illness Cognition Questionnaire (ICQ) Subscale of Work and Life Attitude Scale (WLAS) 105</td>
</tr>
</tbody>
</table>

**OUTLINE OF THE THESIS**

This thesis focuses on evaluating the psychosocial functioning, the emotional well-being and the factors affecting the vocational success and well-being in young adults with a Wajong benefit for a childhood-onset chronic somatic condition while growing up. The general introduction of this thesis is covered in the current chapter (chapter 1). In chapter 2 we assessed the developmental trajectory (course of life) of young adults with a disability benefit as a result of chronic somatic conditions and compared this with that of peers from the general Dutch population. Chapter 3 compares the HRQoL, anxiety and depression of
our study group with that of peers from the general Dutch population. Subsequently, the course of life and the HRQoL of young adult women with a Wajong benefit as a result of Juvenile Idiopathic Arthritis compared with the general Dutch population is described in chapter 4. We examined the illness cognitions (IC) of young adults with Wajong benefits because of chronic health conditions in chapter 5 and examined whether there is an association of IC with Health Related Quality of Life (HRQoL), anxiety and depression in this study population. In chapter 6 we investigate possible differences between the psychosocial development trajectory of young adults with childhood cancer with and without a disability benefit. Finally, we examined the disease characteristics, work characteristics, and work motivation of young adults with disability benefits as a result of chronic conditions and the association of those factors and IC with the likelihood of being employed in chapter 7. This thesis ends with the general discussion in chapter 8 of the main results, a reflection of the methodology, clinical and societal implications and key messages.
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