

TRANSITION

TO

Interventions for young adults
with chronic physical conditions
to support self-management
and work participation

ADULTHOOD



Marjolijn Irene Bal

**Transition to adulthood:
Interventions for young adults with chronic physical
conditions to support self-management and work
participation**

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Transition to Adulthood: Interventions for young adults with chronic physical conditions to support self-management and work participation

Transitie naar volwassenheid: Interventies voor jongvolwassenen met chronisch lichamelijke aandoeningen om zelfmanagement en participatie in werk te ondersteunen

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1

General introduction

GROWING UP WITH A CHRONIC PHYSICAL CONDITION

The numbers of young people with a chronic physical condition are growing worldwide. At the same time, more and more of these young people make the transition into adulthood as a result of the earlier detection of chronic conditions and improvements in medical care [1, 2]. Table 1 presents an overview of percentages of people with chronic physical conditions among young adults (aged between 16-24 years) in the countries of The European Union and the Netherlands. In Europe, 15.7% of the young adults live with a chronic condition, versus 8.9 % of the young adults in the Netherlands [3].

Table 1. Percentages of people with a chronic physical condition in Europe and The Netherlands

	Europe %	The Netherlands %
Working age population (16-64 yr)	29.3	17.8
Young adults (16-24 yr)	15.7	8.9

For every young person, the transition into adulthood requires complex life changes [4, 5]. Young people are expected to become autonomous adults, who live independently from their parents, have followed education and participate in work and society [6, 7]. This transition in itself is difficult, but is extra complicated for young people with chronic physical conditions [4, 8]. These young people have to weigh up the typical developmental tasks of adolescence and young adulthood with the medical challenges posed by the chronic physical condition [8].

In several studies, young people with chronic physical conditions reported limited autonomy and social participation [9-11]. Nevertheless, proper psychosocial development during adolescence and young adulthood is important for adjustment to adult life [6, 9, 10]. Proper psychosocial development means that the young adult learns to deal with symptoms, treatment and the psychosocial consequences of having a chronic physical condition, and thus develops self-management behavior [9]. At each developmental stage during childhood and adolescence new age-appropriate life skills are acquired, which are important for social participation now and in the future [6, 7, 12]. Several studies showed that young adults with chronic physical conditions who have reached fewer developmental milestones in childhood and adolescence, have a limited social participation in adult life [4, 13-15]. The World Health Organization defines these life skills as ‘the abilities for adaptive and positive behavior that enable individuals to deal effectively with the demand and changes of everyday life’[16]. Commonly cited life skills include communication skills, problem-solving skills, and coping skills such as increased self-esteem and the ability to manage emotions and stress [16].

Thus, the transition into adulthood might be challenging for young people with chronic physical conditions. Recent studies showed that these young people are restricted in social participation [9-11]. Because proper psychosocial development is associated prerequisite for successful social participation in (young) adulthood [6,9,10], it is important to incorporate a lifetime perspective on psychosocial care for this group, focusing on self-management and life skill development, and on social participation.

SELF-MANAGEMENT AND LIFE SKILL PROGRAMS FOR YOUNG PEOPLE WITH CHRONIC PHYSICAL CONDITIONS

The WHO recently redefined health as “a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity”[17]. Therefore, it seems suitable to operationalize the comprehensive definition of self-management referring to “dealing with symptoms, treatment, physical and psychosocial consequences of a chronic physical condition” into self-management support [18]. Although it is widely acknowledged that self-management is about dealing with the consequences of the chronic physical condition in activities of daily life, still the self-management support is mostly focused on self-care and medical tasks [19]. Little is known about how self-management – in the sense of the broad definition – is operationalized in self-management interventions [20]. Different conceptualizations of self-management can be found in the literature, leading to very diverse evaluation studies. Consequently, little is clear about the effectiveness and effective components of self-management interventions for young people with chronic physical conditions.

Besides self-management interventions, life skill interventions have been suggested to be appropriate interventions to stimulate the psychosocial development of young people with chronic physical conditions [21-24]. Core components of these interventions are discussions, roleplaying/ behavioral practice of specific skills, modeling, coaching, didactic teaching/instructing, and real-world experiential learning. These interventions are focused on self-perception, assertive behavior, personal relationships, occupational and independent living skills and social, educational, and occupational roles [22]. Although most of the relevant studies demonstrated improvements in life skills, limited significant evidence for effectiveness and effective components was found because of the heterogeneity of interventions, skill focus, and disabilities, and shortcomings of study design [21, 22].

In short, several self-management and life skill interventions are described in the scientific literature. It is not clear, however, how effective these are and what the effective components are.

VOCATIONAL PARTICIPATION AMONG YOUNG ADULTS WITH CHRONIC PHYSICAL CONDITIONS

During the transition to adulthood, finding and maintaining employment is one of the challenges young adults with chronic physical conditions face [25-28]. Being employed is an important developmental milestone, since it provides financial independence and is important for psychological wellbeing, i.e. it structures the day, provides social interaction, and contributes to the development of self-identity [29, 30]. However, young adults with chronic physical conditions experience substantial difficulties in their vocational participation. Frequently reported problems are not being able to handle the workload and task demands, difficulties with travelling to work, limited accessibility of buildings, dealing with prejudices of colleagues and employers, and finding a good balance between work and family life [25, 28, 31-35].

As a result, the employment rates among people with chronic physical conditions lags behind compared to the rates in the general population. In the Netherlands and Europe, respectively 42.1% and 33.9% of young adults with chronic physical disabilities have paid employment, compared to respectively 64.9% and 33.5% of their age peers in the general population (Table 2) [2, 36]. Also in the working age population (16-64 years), the paid employment rates of people with chronic physical conditions are lower than the paid employment rates of the general population of the same age (Table 2) [2, 36]. A chronic physical condition also negatively influences other employment characteristics, such as lower productivity, higher sick leave, higher job turnover, or job loss [37]. Young adults with chronic physical conditions reported barriers for finding and maintaining employment and they would like to have vocational support [27, 38].

Thus, young adults with chronic physical conditions report negative experiences and problems with vocational participation. Healthcare professionals, policy-makers and young adults themselves underline the need for support.

Table 2. Paid employment rates in Europe and The Netherlands

	Europe %	The Netherlands %
Employment rates of people with chronic physical disabilities		
Working age population (16-64 yr)	47.3	42.7
Young adults (16-24 yr)	33.9	42.1
Employment rates of people without chronic physical disabilities		
Working age population (16-64 yr)	66.9	80.1
Young adults (16-24 yr)	33.5	64.9

VOCATIONAL REHABILITATION PROGRAMS FOR YOUNG ADULTS WITH CHRONIC PHYSICAL CONDITIONS

‘At Work’ is a Dutch vocational rehabilitation intervention especially developed for young adults (aged between 16-27 years) with chronic physical conditions who have problems finding competitive employment after finishing post-secondary education [39, 40]. This multidisciplinary intervention – provided by a job coach and an occupational therapist – consists of six group sessions and an individual one-year coaching trajectory. The intervention aims (a) to improve young adults’ self-efficacy for dealing with disease and work-related problems and life skills, and (b) to provide support for vocational participation. Verhoef and colleagues in 2013 systematically evaluated the intervention and found that 8 out of 12 participants were employed two years after the start of the intervention; the authors concluded, therefore, that the intervention is feasible [39].

Another vocational intervention for young people with chronic physical conditions mainly addresses the development of life skills in combination with support in finding temporary jobs while they are still receiving education [21]. The aim of the program is to explore career interests and to develop essential life skills, such as social communication skills and problem solving skills. It also offers workshops in which participants’ life skills are trained, as well as individual assessments, coaching sessions, and peer support meetings. Community placement matches were facilitated by an occupational therapist and jobcoach. Insights into the effectiveness of this program, evaluated with a quantitative controlled study design, are lacking.

To our knowledge, in the Netherlands, three initiatives other than the ‘At Work’ program, support vocational participation among young adults with a chronic physical condition, i.e. ‘Emma at work’ [41], ‘Brains4U [42], and The Class [43]. Outcomes of these other three initiatives have not yet been systematically evaluated in a controlled trial.

In short, 'At Work' is the only intervention specifically focused on young adults with chronic physical conditions who experience problems in finding employment job after finishing post-secondary education. More significant evidence is needed on the effectiveness of vocational support to this specific group.

THIS THESIS

Study aim and research questions

Part I

As described above, young adults with chronic physical conditions are at risk for delay in psychosocial development [9-11] and need support on life skills development and self-management behavior. There is no clear insight, however, in how effective the current self-management support program are, and what constitute effective components [19, 44, 45]. The group of young people receiving disability benefits is specific subgroup of young people who have a less favorable developmental trajectory while growing up as compared to healthy age-mates [4, 13, 46]. Little is known, however, about why and how psychosocial development might differ between young people with chronic physical conditions who apply for disability benefits and those who do not [14]. Moreover, potential predictors for disability benefit applications are not known. Closing these knowledge gaps may help to identify a target group that will benefit from timely support on self-management or life skills development and (pre)-vocational participation. Therefore, the research aim and questions of the first part of this thesis were:

I: To explore the content, effective components, and effectiveness of self-management support programs; to identify a subgroup of young people with chronic physical conditions vulnerable for a delay in psychosocial development.

- a) What are characteristics, contents, and expected outcomes of self-management interventions for young people with chronic physical conditions?
- b) What is the effectiveness and effective intervention components of self-management support for young people with chronic physical conditions?
- c) What are predictors during adolescence for the application on disability benefits during young adulthood and how does the psychosocial development differ between young people with chronic physical conditions who apply for disability benefits and those who do not.

Part II

Nazarov and colleagues (2019) reviewed the contents of interventions aimed to support job retention and return to work among workers with chronic conditions, and found that insights into the effectiveness of these vocational rehabilitation interventions are lacking [47]. Of note, 'At Work' is the only vocational rehabilitation program described in the literature that has been specially developed for young adults with chronic physical conditions who experienced problems with finding competitive employment after finishing their post-secondary education [39, 40]. Significance evidence of the perceived working mechanisms and effectiveness of this (Dutch) vocational rehabilitation program is lacking. Therefore, the research aim and questions were:

II: To study effectiveness of a) vocational rehabilitation programs in general; and b) a specific Dutch vocational rehabilitation program ('At Work') focused on young adults with chronic physical conditions

- a) What are the characteristics and effectiveness of vocational rehabilitation programs for people with chronic physical conditions?
- b) How do former intervention participants perceive the beneficial attributes of the 'At Work' program?
- c) Has the paid employment rate of former 'At Work' participants changed over time, and does it equal the paid employment rate of a national reference population of young adults with chronic physical conditions?
- d) What is the effect of the 'At Work' program on participants' self-efficacy for dealing with work-related problems and paid employment?

Study population

Our research was focused on young people aged 16–27 years with chronic physical conditions¹. Since young adults with different chronic physical conditions may encounter comparable challenges entering the labour market, and face similar adaptive tasks irrespective of type of condition [48, 49], the vocational rehabilitation program 'At Work' program, and thus our study sample, included a heterogeneous sample of young adults with a broad range of chronic physical conditions.

1 The study samples in this research included young people with chronic physical conditions – either congenital or acquired in childhood. In this thesis, a **chronic condition** is defined according to the definition proposed by Mokkink and colleagues (2008) [53]: "A disease or condition is considered to be a chronic condition in childhood if: (1) it occurs in children aged 0 up to 18 years; (2) the diagnosis is based on medical scientific knowledge and can be established using reproducible and valid methods or instruments according to professional standards; (3) it is not (yet) curable or, for mental health conditions, if it is highly resistant to treatment and (4) it has been present for longer than three months or it will, very probably, last longer than three months, or it has occurred three times or more during the past year and will probably reoccur".

Study context

At the time of our research, young adults in the Netherlands who are partially or fully incapable of working due to childhood onset of a chronic condition could be entitled to a benefit under the Wajong Act (the Disablement Assistance Act for Handicapped Young Persons) [50]. Applicants needed to be at least 25% occupationally disabled before their 17th birthday or have become occupationally disabled during their education. The disability benefit consists of (supplementary) income support, and support in finding and sustaining employment for those who are fit to work [4, 50]. Disability benefit recipients reported low employment rates: about 25% of them were employed, whereas 60% of all recipients were found fit to work [51]. Also in other countries, lower employment rates are reported for benefit recipients compared to non-recipients [52].

Outline of this thesis

We systemically reviewed international scientific literature on the content, effective components and effectiveness of self-management interventions and vocational rehabilitation programs. **Chapter 2** considers the underlying theories, content, and intervention formats of self-management support of these young people. In **Chapter 3**, the results of a systematic review on the effectiveness, and effective intervention components of these self-management interventions are described. Then, we investigated the association between characteristics during adolescence and the likelihood of receiving disability benefits in young adulthood. In addition, differences between recipients and non-recipients of benefits on participation and psychosocial outcomes in young adulthood were studied (**Chapter 4**). The results of a systematic review on the characteristics and effectiveness of interventions aimed to improve the vocational participation of people with chronic physical conditions are described in **Chapter 5**. Only the 'At Work' program is described in previous literature as a specific vocational rehabilitation program for young adults with chronic physical conditions who experience problems with finding competitive employment after finishing post-secondary education. Significant evidence on perceived beneficial attributes of the intervention and effectiveness is lacking. In **Chapter 6**, intervention participants' perception of beneficial attributes of the 'At Work' program were evaluated. **Chapter 7** considers the development of intervention participants' employment rates ($n=90$) compared to national reference data. Thereafter, the effectiveness of the 'At Work' program on self-efficacy for dealing with disease and work-related problems and vocational participation was evaluated, using a multicenter controlled trial design (**Chapter 8**). Lastly, **chapter 9** includes the general discussion of main findings of this thesis, a reflection on methodological considerations, discussion of practical implications, and recommendations for further research.

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diseases and health conditions in childhood (0-18 years of age): national consensus in the Netherlands. *Eur J Pediatr*, 167(12), 1441-7

2

Self-management interventions for young people with chronic conditions: A systematic overview

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ABSTRACT

Objective: To provide a systematic overview of self-management interventions (SMI) for young people with chronic conditions with respect to content, formats, theories, and evaluated outcomes.

Methods: Embase, Medline, PsycINFO, Web-of-Science, CINAHL, and Cochrane were searched. Reviews' reference lists were scrutinized. Selected studies were: Original research articles in English published between 2003 and March 2014; about the evaluation of SMI for 7 to 25-year-olds with somatic chronic conditions/physical disabilities; with clear outcomes and intervention descriptions. The classification of medical, role and emotion management served to review content. Formats, theories, and evaluated outcomes were summarized.

Results: 86 studies were reviewed. Most aimed at medical management and were unclear about theoretical bases. Although a variety of outcomes was evaluated and the distribution over self-management domains was quite unpredictable, outcomes conceptually related to specific content. A content-based framework for the evaluation of self-management interventions is presented.

Conclusions and practice implications: SMI relate to self-management tasks and skill-building. Yet, conceptualizations of self-management support often remained unclear and content focuses predominantly on the medical domain, neglecting psycho-social challenges for chronically ill young people. Future evaluations should match outcomes/themes to content and characteristics. Our framework and overview of SMI characteristics and outcomes may assist clinicians in providing self-management support.

INTRODUCTION

Worldwide, the number of young people living with a chronic condition or with special health care needs is growing. In the USA, the 2009-2010 National Survey of Children with Special Health Care Needs showed that 15.1% of all under 17-years-olds fell in this category [1]. In the Netherlands, the most recent estimations are 14% of all under 18-year-olds [2] and 11% of all under 25-year-olds [3].

Chronic illness affects young people in many ways during their transition to adulthood and adult care [4,5]. Supporting them to develop independence and self-management skills is therefore a key task of healthcare professionals. For that matter, self-management support is considered an integral part of healthcare for all people with chronic conditions [6-8]. The WHO definition of health was even redefined as “the ability to adapt and self-manage in the face of social, physical, and emotional challenges” [9].

Living with a chronic condition is an “ongoing process of inner negotiation” between social and medical needs [10] or what is described as shifting between the illness-on-the-foreground and wellness-on-the-foreground perspective [11]. Self-management therefore has been defined as “the individual’s ability to manage the symptoms and the consequences of living with a chronic condition, including treatment, physical, social, and lifestyle changes” [12]. Note, however, that self-management is not restricted to one’s individual ability, especially not in pediatrics where parents tend to play a key role. Adding the phrase “ [...] in conjunction with family, community, and healthcare professionals [...]” [13] seems to present a more complete picture. This holistic view accounts for the three tasks involved in self-management: medical management (*re.* treatment), role management (*re.* social participation), and emotion or identity management (*re.* emotional consequences of being ill) [14]. Young people with chronic conditions have to learn these tasks, and in supporting them we must take their developmental transition into account [15].

Various self-management interventions (SMI) for the chronically ill are available, but their effectiveness is not clear [16,17]. This is even more pertinent to SMI in pediatric care [16,18,19]. Newman and colleagues (2009) emphasize that a theory-based approach is needed to evaluate complex SMI, and recommend a more systematic comparison of different types of SMI [20]. Recent studies on SMI for people with chronic conditions in general [17,21] and for young people with physical disabilities [19] endorse this view, and it is recommended to standardize SMI evaluation by using a core set of outcomes [19,22].

We reviewed and systematically compared the characteristics and content of offered SMI for young people (7-25 years) with chronic conditions, their theoretical foundations, if any, and the evaluated outcomes. Based on the results we present content-related outcome measures for the evaluation of different types of self-management interventions.

METHODS

Study design

A systematic overview, defined by Grant and Booth [23], as a “summary of the literature that attempts to survey the literature and describe its characteristics” was applied. This allows for a systematic comparison of SMI and outcome measures used in evaluation studies. Methodological characteristics according to the ‘Search, Appraisal, Synthesis and Analysis’ (SALSA) framework [23] are: comprehensive searching, quality assessment, narrative synthesis with tabular features, and thematic analysis. The review process was guided by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement [24].

Search strategy

The search strategy employed variations and Boolean connections (AND, OR) of the following terms: self-management, children, adolescents, young adults, chronic illness, and intervention. Relevant variations were derived from database thesauruses and relevant review articles (i.e. childhood, youth, chronic disease, physical disability, program etc.). Six health-related databases were searched: Embase, Medline, PsycINFO, Web-of-Science, CINAHL, and Cochrane. An information specialist helped define the final search strategies, employing a combination of free-text and thesaurus terms. The strategy used in Embase is presented in Box 1. Two researchers (JS, MB) supplemented the database searches by scrutinizing relevant reviews’ references for additional relevant publications.

Inclusion criteria

- Study types: only original research articles in English language published from 2003 to March 2014. No restrictions were placed on study design.
- Participants: young people (aged 7–25 years) with somatic chronic conditions or physical disability.
- Interventions: studies focusing on the evaluation of an SMI and describing the SMI or referring to previous description(s) of the intervention.

- **Outcome measures:** No restrictions were placed on the type of outcome measures, as this was our main interest. However, outcome measures needed to be clearly defined.

Studies had to meet all inclusion criteria to be included for further analysis. Furthermore, the term ‘children’ is used for young people aged 7-12 years, the term ‘adolescents’ is used for the age group of 13-18 years, and the term ‘young adults’ is used for those aged 19-25 years.

Box 1. Search strategy in Embase

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((('self care'/de OR 'self medication'/de OR 'self help'/de OR 'drug self administration '/de OR (((self OR shared) NEAR/3 (manag* OR care* OR medicat* OR efficac* OR help*)))ab,ti) OR (((('coping behavior'/exp OR 'health education'/de OR 'patient education'/de OR emotion/de OR emotionality/de) AND ('intervention study'/de OR psychotherapy/exp OR 'program development'/de)) OR (psychotherap* OR ((coping OR cope OR cognitiv* OR behavior* OR emotion* OR education* OR psychologic*) NEAR/6 (therap* OR interven* OR program*)))ab,ti)) AND ('chronic disease'/de OR 'genetic and familial disorders'/exp OR 'congenital disorder'/exp OR 'disabled person'/de OR 'handicapped child'/de OR disability/exp OR (((chronic* OR longterm OR 'long term' OR 'end stage' OR endstage* OR degenerat* OR persisten* OR genetic* OR familial* OR congenit*) NEAR/3 (ill* OR disease* OR condition* OR disorder*)) OR (physic* NEAR/3 (handicap* OR disab* OR challeng*)))de,ab,ti) AND (child/exp OR adolescent/exp OR adolescence/exp OR 'child health care'/de OR 'child care'/de OR 'child hospitalization'/de OR 'handicapped child'/de OR (young OR youth OR child* OR adolescen* OR teenage* OR teen OR teens OR juvenile*):ab,ti) AND ('comparative effectiveness'/de OR 'clinical effectiveness'/de OR evaluation/de OR 'self evaluation'/de OR (effectiv* OR evaluat*):ab,ti)
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Selection, quality assessment, and data extraction

Retrieved records (n = 5908) were imported into Endnote®. Two reviewers (JS, MB) independently selected eligible studies from both title and abstract and categorized them into: include, exclude or not clear. Any discrepancies were resolved, and decisions were made on the ‘not clear’ category. Full texts of all agreed-upon articles (n = 444) were retrieved. The two reviewers decided on final inclusion of articles based on the full text, resulting in 103 publications. The selection process is presented in Figure 1. Three reviewers (JS, MB, PR) assessed methodological quality of randomized controlled trials and cohort studies with methodology checklists of the Scottish Intercollegiate Guidelines Network (SIGN) [25]. For qualitative studies the ‘Consolidated criteria for reporting qualitative research’ (COREQ) checklist [26] was used. Any discrepancies were resolved by discussion. Seventeen studies were excluded because outcome measures were not clear, leaving 86 studies. Two reviewers (JS, MB) extracted data on study design, study sample, type and content of interventions, settings of interventions, interventionists, theoretical basis, and outcome measures. Data were recorded in an electronic extraction form.

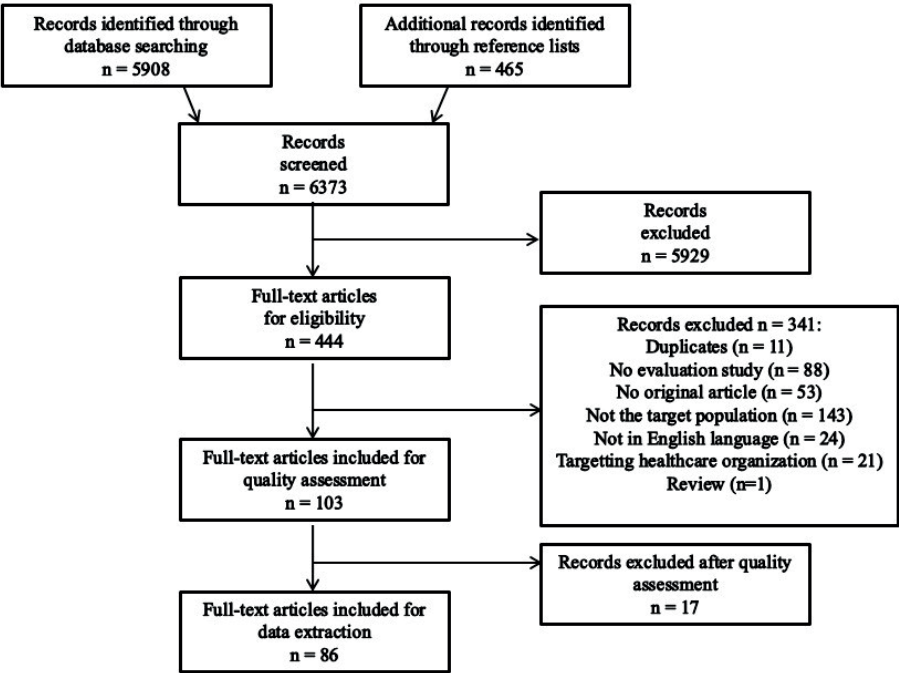


Figure 1. Selection process

Analysis

General study characteristics were summarized, i.e. study country, chronic conditions addressed and study designs, as well as SMI characteristics, i.e. the modes, formats, elements and settings of SMI and professionals involved. Lorig and Holman’s classification of domains of self-management [14] served as a framework to review the content of SMI. Interventions could be aiming at medical management, role management, emotion management or a combination thereof. Further analysis included comparisons of theories underlying SMI per self-management domain. Finally, evaluated outcome measures were inventoried and linked to the content of SMI. On the premise that certain outcome measures logically relate to specific content of SMI, one reviewer (JS) linked all outcome measures to the content descriptions. Another reviewer (MB) checked this to enhance validity of this analysis.

RESULTS

General study characteristics (n=86)

- Countries: Most studies hailed from the USA (n=51), followed by the Netherlands (n=8), the UK (n=7), Australia (n=4), Canada (n=3), Germany (n=3), Hungary (n=2), Taiwan (n=2), Austria (n=1), China (n=1), Denmark (n=1), France (n=1), Haiti (n=1), and Norway (n=1).
- Chronic conditions: Most studies targeted asthma (n=18), followed by diabetes (n=16). Six studies targeted several chronic conditions (Table 1).
- Study designs: All but nine studies had fully quantitative study designs. Forty-five of them were randomized controlled trials, 29 were cohort studies and 3 were cross-sectional studies. Three studies had fully qualitative study designs, while five were mixed-methods studies and one was a case study. Twenty-six studies (30.2%) were classified as pilot evaluations.
- Interventions: A total of 81 different interventions were reviewed, because different studies evaluated the same intervention with different outcome measures ([27] and [28]; [111] and [112]; [93] and [94] and [95]; [75] and [77]).

Intervention characteristics (n=81)

Interventions were either applied at individual level (n=39; 48.1%), at group level (n=34; 42.0%) or both (n=8; 9.9%). Most interventions included educational and/or skills training sessions (n=35; 43.2%) or telemedicine systems (n=14; 17.3%). Intervention formats and elements are summarized in Table 2. In 20 interventions (24.7%), parents were included as participants. These interventions often considered educational and/or skills training and most included both separate and joint sessions. Three interventions (3.7%) offered joint sessions only, while seven interventions (8.6%) offered separate but parallel sessions for parents and their children. Intervention settings were camping sites (n=10; 12.4%), inpatient or outpatient clinics (n=35; 43.2%), home or public environments (n=13; 16.0%), school (n=9; 11.1%), or online (n=10; 12.4%). Settings were not exclusive for the formats of interventions. Four studies (4.9%) did not detail the settings.

Interventionists included pediatricians, nurses, physiotherapists, occupational therapists, psychologists, social workers, pedagogues, dietitians, job coaches, and speech pathologists. In some cases, interns or research assistants were additionally available. Occasionally, the whole healthcare team was involved. Twenty-two studies (27.2%) lacked this information. See Appendix A for an overview of general study characteristics and intervention characteristics per study.

Table 1 Studies by chronic condition (n=86)

Chronic condition	References	No. (%)
Asthma	[26-43]	18 (20.9)
Diabetes	[44-59]	16 (18.6)
Cancer	[60-64]	5 (5.8)
Chronic fatigue syndrome	[65]	1 (1.2)
Chronic condition (various)	[66-71]	6 (7.0)
Chronic pain	[72-76]	5 (5.8)
Chronic respiratory condition	[77]	1 (1.2)
Cystic Fibrosis	[78-81]	4 (4.7)
Eczema (atopic dermatitis)	[82]	1 (1.2)
End-stage renal disease	[83-85]	3 (3.5)
Epilepsy	[86]	1 (1.2)
Heart disease	[87]	1 (1.2)
Hiv	[88,89]	2 (2.3)
Inflammatory bowel disease	[90]	1 (1.2)
Ichthyosis	[91]	1 (1.2)
Juvenile Fibromyalgia	[92-94]	3 (3.5)
Juvenile Idiopathic Arthritis	[95-98]	4 (4.7)
Migraine	[99]	1 (1.2)
Phenylketonuria	[100]	1 (1.2)
Physical disability	[101-103]	3 (3.5)
Sickle cell disease	[104-108]	5 (5.8)
Spina bifida	[109-111]	3 (3.5)

Medical, role and emotion management: Content of self-management interventions

The content of interventions includes the actual themes, topics, issues or specific skills discussed, reviewed or practiced during the interventions. Content is categorized by the domains of self-management [12] in Table 3. Many interventions (46.2%) were solely aimed at medical management; some considered role management (6.4%) or emotion management (2.6%) alone. Others addressed multiple domains, See Figure 2.

Table 2. Formats and elements of self-management interventions according to mode

Modes	Formats	Elements
Individual	Educational sessions (with or without parents) or written materials	- Informational (comic) books and videos - Daily diaries or notebooks (with or without rewards) - Homework assignments (written or skills practice) or workbook - Check-in or booster telephone calls by interventionist - Role reversal (between educator and the one(s) being educated)
	Motivational interviewing sessions	- Awareness building - Problem solving - Goal setting
	(Skills) training sessions	- Symptom treatment (e.g. relaxation techniques or pain provocation technique)
	Cognitive behavioral therapy sessions (some of them with parents)	- Educational and skills training - Instructions for home practice
	Family sessions	- Written materials - Responsibility-sharing plan - Family discussions (with conflict resolution) - Problem solving training - Communication training - Homework assignments (behavior)
	Telemedicine system (e.g. through personal devices, text-messaging, websites, or web-based systems)	- Monitoring through daily diaries - Overview of (trends in) disease-specific outcomes - Individualized feedback - Reminders or cueing - Social media communication or online discussion board - ‘Gamification’ (with feedback or rewards), role-playing or knowledge quizzes - Goal-setting or action plans - Information messages, animated lessons or tips - Skills training - Modules with homework - Possibility to contact healthcare provider
	CD-ROM	- Educational modules - Active coping plan - ‘Gamification’ with feedback
	Peer-support (e.g. befriending program)	- Mentorship
	Individual (transition) plan	- Age and developmentally appropriate information resources - Goal-setting

Table 2. Formats and elements of self-management interventions according to mode (continued)

Modes	Formats	Elements
Group	Cognitive behavioral therapy sessions	- Fun activities and games or role-playing - Homework (skills practice) - Involvement of parents as coaches - Goal-setting
	Art therapy sessions	- Discussion of weekly topics - Art making - Discussing art and related feelings
	Camping programs	- Traditional camping activities (e.g. horse riding, boating, arts etc.) - Disease specific activities (e.g. educational sessions, support groups, discussions, problem solving, role-playing, knowledge-testing games)
	Skills training or workshop	- Goal assessment and goal-setting - Drafting action or transition plans - Practicing strategies for goal achievement (e.g. through role-playing, coaching, use of audio-visual aids, accessing the Internet etc.)
	Educational and/or support sessions	- Informational videos, (coloring) books, written information, educational stories - Didactic presentations - Question and answer sessions - Discussions and problem solving - Homework assignments, exercise books and skills practice - Self-monitoring with contingency management - Self-management plans - Devices for self-monitoring (e.g. peak flow meter)
	Family sessions (parallel but separate groups for children and parents; in some cases one mixed session)	- Play therapy, narrative therapy or role play - Relaxation training - Group work - Social support - Training in coping strategies - Homework (practice skills)
	School program (with continued phone contact)	- Didactic presentation about the disease - Peer education

Table 3. Content of interventions categorized by the domains of self-management^a

Domains ^b	Content of interventions	References
Medical management	Disease-specific:	
	1. Understanding the disease	[27, 30,
	2. Understanding (the necessity of) medication and treatment regimen; understanding side effects; adherence	31-50, 52-57, 59, 60,
	3. About the use of specific treatment devices or techniques (e.g. peak flow meter for asthma)	63, 64, 66, 73-76, 79,
	4. Dealing with symptoms	82-85, 87,
	5. Drafting an individualized care plan	90-93, 96,
	6. Self-monitoring of clinical outcomes	100, 101, 105-109]
	General:	
	7. Accessing healthcare	[33, 39, 44,
	8. Communication with healthcare professionals	47, 50, 54,
	9. Managing doctor visits	65, 76, 82,
	10. Coping with hospitalizations	84, 85, 89,
	11. Goals and dreams for the future related to health and healthcare (transition)	92, 96, 98,
	12. Child-parent sharing / teamwork related to disease-specific medical management	103, 105, 110]
	13. Knowing where to find specific information about the disease	
	14. Knowing when to ask for (medical) help	
	15. Risk behavior (e.g. unsafe sex or drug and alcohol abuse)	
Role management	1. Social initiation and friendship making; social networks; family and romantic relationships	[27, 29, 33, 39, 47, 51,
	2. Managing teasing and bullying; conflict resolution	57, 59, 61-63, 65-67,
	3. Participating in normal social activities; keeping up with peers; Internet and social media	69, 71, 72,
	4. Goals and dreams for the future related to school, work, community, living, housing, recreation and leisure (looking ahead); school issues	76, 81, 82, 86, 87, 89,
	5. Romantic relationships and sexuality	92, 96-99,
	6. Explaining the condition to others (disclosure); educating peers	102-104,
	7. Setting (life) goals and becoming assertive; growing up	107, 110,
	8. Communication and social problem solving (sometimes within families); organizational skills	111]
	9. Independent living; traveling/staying abroad	
	10. Social rights and benefits	
Emotion management	1. Self-confidence or self-esteem building; developing a positive body image; body esteem	[29, 40, 42, 47, 49, 51,
	2. Self-appreciation; enhancing hope; enhancing self-efficacy	59, 61, 62,
	3. Empathy; fear-related thinking;	65-67, 70,
	4. Feelings related to condition; sharing of feelings and experiences	71, 74, 76,
	5. Accepting condition; self-reflection	80, 83, 86,
	6. Healthy expressions of anger and transforming or managing anger	87, 96, 99,
	7. Helpful / positive thoughts; stress management	100, 103,
	8. Decreasing negative thoughts	104, 111]
	9. Decreasing stress and boredom; decreasing social isolation	
	10. Spirituality	
	Emotions	

^aNumber of studies is 78, three studies were unclear about the content of the intervention: [58, 68, 88]^bAccording to the model of Lorig & Holman (2003) [14]

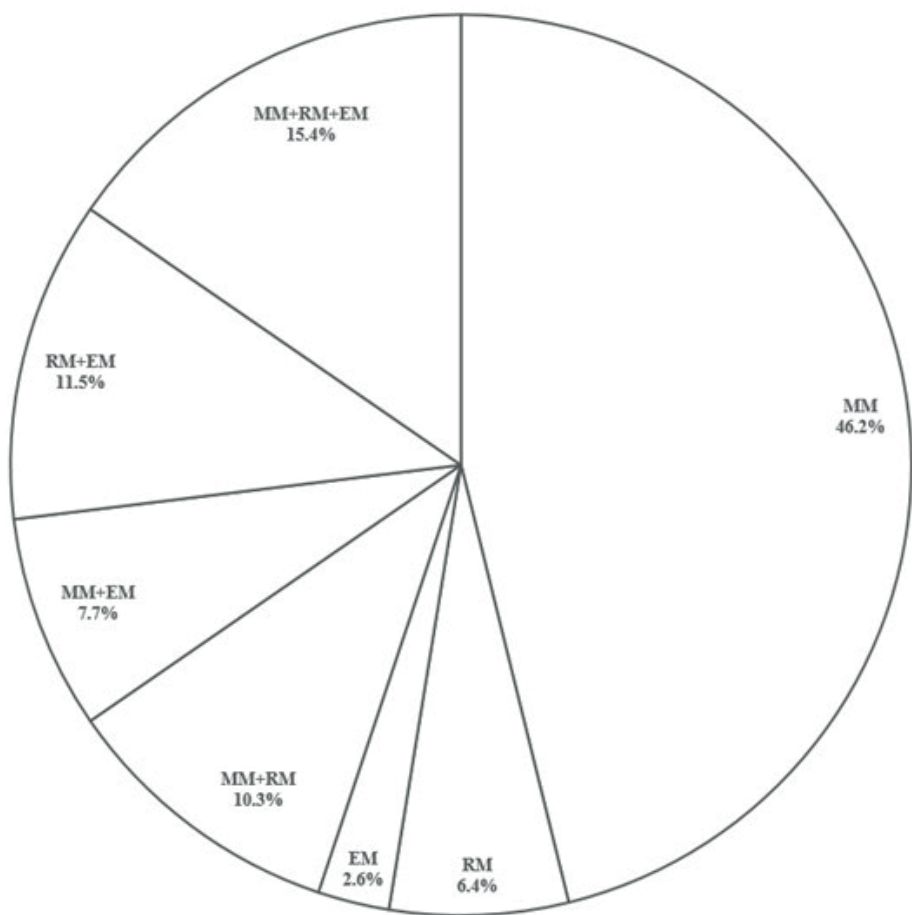


Figure 2. Distribution of interventions (n=68) over (combinations of) self-management domains*
*MM - medical management, RM - role management, EM - emotion management

Medical management was either disease-specific or of a more general nature. The former refers to tasks or topics associated with or related to a specific diagnosis, e.g., self-monitoring of blood glucose values in diabetes. This type of content is not exchangeable between interventions, e.g., education on treatment of cystic fibrosis is not useful for renal transplant patients. General medical management refers to health and healthcare related tasks irrespective of diagnosis. For instance, accessing healthcare, but also child-parent sharing or teamwork related to medical management tasks.

Role management referred to tasks or topics on domains related to social participation, such as communicating, decision-making, assertiveness, and keeping up with peers. Domains are school, work, community, living, housing, recreation, sports and leisure,

relationships and sexuality. A major focus is on peer relationships and disclosure of the condition in social environments.

Emotion (or identity) management referred to the young person's feelings and intrinsic characteristics. Topics covered are building self-confidence, developing a positive body image, self-appreciation, maintaining positive thinking, stress management, but also acceptance of the condition.

The content of interventions was not specifically linked to certain modes, formats, elements or settings of SMI. In general, interventionists were not exclusive for content of interventions, although occasionally specific interventionists were included, e.g., a sexologist. See Appendix A for the classifications of self-management domains per study.

Self-management interventions for different age groups

Most interventions targeted 12 to 18-year-olds ($n=36$; 44.4%) or 7 to 11-year-olds ($n=23$; 28.4%). Only five SMI (6.2%) targeted over 18-year-olds. For the rest, age groups overlapped. Formats and classification of self-management domains did not seem to be related to specific age groups, but content or themes obviously were not applicable to the whole age range. For example, an intervention classified as targeting both role and emotion management for children (mean age 10 years) targeted communication and social problem solving in general [50], while for young people (mean age 20 years) such an intervention targeted the social subtheme of intimate relationships [64]. Another theme specific for older age groups is vocational participation. Two interventions aimed at the whole age range (7 to 25 years) addressed medical management and self-monitoring through daily diaries, respectively.

Conceptualization of self-management: Theoretical bases of self-management interventions

Fifty-five studies (67.9%) either failed to state whether the interventions were based on a theory ($n=48$) or, if they did so, did not specify the theoretical base ($n=7$). Of the other studies, most referred to learning theories like Bandura's (cognitive) social learning theory or cognitive behavioral theory (Table 4). A theoretical base was mostly mentioned in relation to interventions targeting medical management alone, while only one of the studies evaluating role management interventions mentioned a theoretical base. In general, neither the content of interventions nor intervention characteristics were specific for a certain theoretical base.

Table 4. Theoretical bases of self-management interventions, no. (%)

Theoretical base	Number of interventions (n=26)	References
(Cognitive) social learning theory	10 (38.5)	[29, 31, 48, 51, 59, 65, 75, 76, 79, 89]
Cognitive behavioral theory	9 (34.6)	[64, 66, 70, 74, 75, 91, 93, 106, 109]
Health Belief Model	2 (7.7)	[35, 85]
Prochaska's transtheoretical model	1 (3.8)	[35]
Self-regulation model of health and illness	1 (3.8)	[65]
Transactional model of stress	1 (3.8)	[40]
Orem's self-care deficit theory of nursing	2 (7.7)	[39, 44]
Game-playing and health theory	1 (3.8)	[108]
Flirt Model	1 (3.8)	[67]
Self-confrontation	1 (3.8)	[99]
Model of Human Occupation	1 (3.8)	[104]

Evaluating self-management interventions: Measured outcomes

Interventions were evaluated on a wide variety of outcomes, primarily health outcomes (61.5%), health-related quality of life (HRQoL) (35.9%), and knowledge about the disease and/or treatment (29.5%) (Table 5).

Interventions solely aimed at medical management (n=36) were evaluated on all outcome measures except psychosocial functioning, and support by others. Of the five interventions solely aimed at role management, two were evaluated only on health outcomes, two on psychosocial functioning and one on social participation. One of the two emotion management intervention studies evaluated knowledge of disease and/or treatment, and the other social participation (Table 5). None of the outcomes or groups of outcomes could be related to one particular type of intervention and the distribution over self-management domains or combinations of self-management domains was quite unpredictable. Appendix A presents an overview of outcome measures per study and the groups of outcomes.

Linking content and outcomes: A content-based evaluation framework

Regarding the content of interventions (Table 2), certain content logically relates to groups of outcomes or themes. If, for example, ‘understanding of the disease’ and ‘adherence’ is addressed, it would seem logical to evaluate intervention effectiveness from improved knowledge, clinical outcomes and self-reported adherence rather than from psychological outcomes such as depressive symptoms or anxiety. Grounded on this premise, a conceptual content-based measurement framework for the selection of outcome measures in the evaluation of SMI is presented in Figure 3. The outcome mea-

Table 5. Outcomes used in the evaluation studies distributed over (combinations of) self-management domains

(Combined) domains of self-management ^a No. (% of total studies ^b)	MM n=36 (46.2)	RM n=5 (6.4)	EM n=2 (2.6)	MM+RM n=8 (10.3)	MM+EM n=6 (7.7)	RM+EM n=9 (11.5)	MM+RM+EM n=12 (15.4)	Total ^b n=78
Groups of outcome constructs or themes ^c								
Health outcomes	27 (75.0)	2 (40.0)		4 (50.0)	5 (83.3)	3 (33.3)	7 (58.3)	48(61.5)
Health-related quality of life	13 (36.1)			5 (62.5)	1 (16.7)	4 (44.4)	5 (41.7)	28(35.9)
Knowledge of disease / treatment	12 (33.3)		1 (50.0)	6 (75.0)		2 (22.2)	2 (16.7)	23 (29.5)
Psychological outcomes	7 (19.4)			1 (12.5)	1 (16.7)	5 (55.6)	2 (16.7)	16 (20.5)
Self-efficacy	8 (22.2)			3 (37.5)		2 (22.2)	2 (16.7)	15 (19.2)
Vocational participation	5 (13.8)			2 (25.0)	1 (16.7)	2 (22.2)	2 (16.7)	12 (15.4)
Social participation	2 (5.6)	1 (20.0)	1 (50.0)	1 (12.5)		5 (55.6)	2 (16.7)	12 (15.4)
Coping	1 (2.8)			1 (12.5)	2 (33.3)	3 (33.3)	1 (8.3)	8 (10.3)
Self-care	2 (5.6)			3 (37.5)			2 (16.7)	7 (9.0)
Psychosocial functioning		2 (40.0)			1 (16.7)	1 (11.1)	2 (16.7)	6 (7.7)
Family involvement or conflict (related to disease-related management tasks)	4 (11.1)			1 (12.5)		2 (22.2)		7 (9.0)
Sense of control	1 (2.8)			2 (25.0)				3 (3.8)
Attitudes towards illness	2 (5.6)			1 (12.5)		1 (11.1)		3(3.8)
Self-perception of competencies	1 (3.1)			1 (14.3)				2 (2.6)
Problem solving	2 (6.3)							2 (2.6)
Support by others			1(50.0)				1 (9.1)	2 (2.6)

^aAccording to the model of Lorig & Holman (2003) [14]: MM = medical management, RM = role management, EM = emotion management

^bNumber of studies is 78, three studies were unclear about the content of the intervention: [58, 68, 88]

^cOnly measured in young people (e.g., no parent proxy measures)

asures correspond to the numbered content descriptions in Table 3. The only outcome related to all three domains was HRQoL.

DISCUSSION AND CONCLUSION

The focus of today's self-management support

This review revealed that most interventions for young people represented in the literature solely aim at medical management, like interventions for adults [17,113,114]. This is not surprising, because medical tasks form the very core of healthcare. Moreover, these tasks represent common ground for healthcare professionals and people with

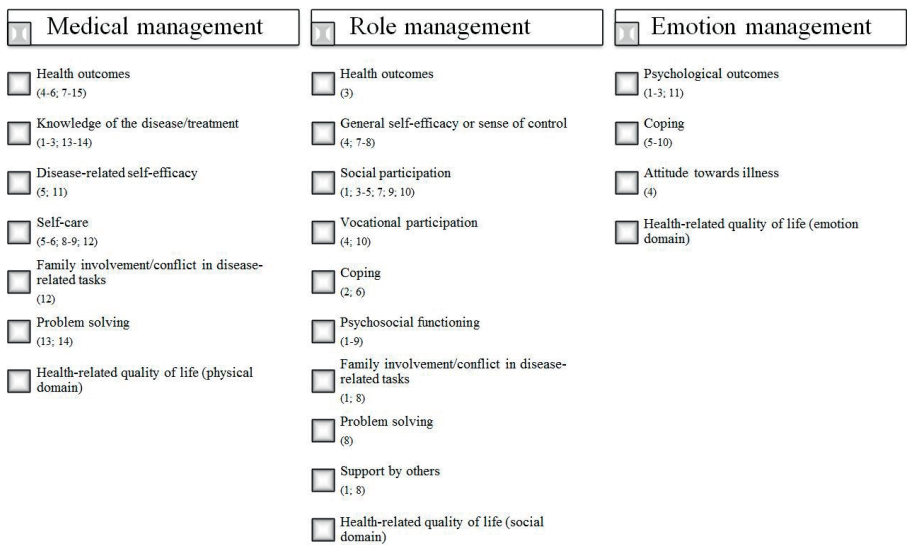


Figure 3 A content-based framework for the selection of (groups of) outcome measures*

*The numbers presented next to the outcomes correspond to specific content in Table 3.

chronic conditions, since medical consultations without fail will address symptoms and treatments. This may also explain why very few interventions address role management or emotion management alone. Still, the fact that 44% of interventions aim at multiple domains indicates a shift in focus of today’s self-management support for young people with chronic conditions. Healthcare professionals nevertheless are challenged to pay more attention to role management and emotion management.

Six self-management skills match the tasks of medical, role and emotion management: “problem solving, decision making, resource utilization, the formation of a patient-provider partnership, action planning, and self-tailoring” [14]. Several SMI indeed were directed at developing such skills, e.g., drawing up an action plan. SMI content also seems to match self-management needs of people with chronic conditions, addressing the following processes: ‘focusing on illness needs’, ‘activating resources’, and ‘living with a chronic illness’ [21]. The first is addressed in, for example, SMI aiming to deal with symptoms, the second in SMI helping young people realize when and how to ask support.

However, the above-mentioned processes basically reflect experiences of adult patients. Additional developmental processes or factors will relate to young people’s self-management processes as well [115], such as ‘determining health needs’ and ‘communication with the medical team’, processes that have been incorporated in the

Pediatric Self-management Model [15]. Several SMI indeed target such processes, albeit the Pediatric Self-management Model seems to more narrowly focus on medical management. Young people have to learn to balance or “articulate” [116] self-management tasks, which their parents use to be responsible for. Parental involvement can either hinder or facilitate adolescents’ development of self-management [117], and professionals and researchers should be aware of this [15,117]. Some SMI involved parents in the intervention or assessed family interaction or conflict. However, the notion that social context deserves attention when researching self-management, has only recently gained more attention [14,17,19,117-120].

The conceptualization of self-management support

For most of the interventions a theoretical base was not provided, which was also found in other reviews of SMI for both adults and young people [16-18]. The studies that did mention a theoretical base often referred to social learning and cognitive behavioral theories which were also found to underlie SMI for adults [16,17]. Social learning theory argues that people learn from others and in general aims at enhancing self-efficacy [121], while employing an “experiential” approach to self-management [17]. In this view, self-management refers to learning about and believing in yourself, and self-management support facilitates environments that allow to ‘learn from others’ and gain ‘mastery experiences’. On the other hand, cognitive behavioral theory aims to change thoughts and attitudes and ultimately behavior [122], and from this point of view self-management support might be targeted at behavior thought to be beneficial from a medical perspective. In this light, it could represent a more “authoritative” approach to self-management [17]. The different theoretical bases thus represent different views on self-management. For young people, the experiential approach seems more appealing, as telling them what to do is less effective. Young people tend to weigh medical advantages against social disadvantages [4]. Moreover, self-assurance would form a firm basis for healthy behavior [115].

Evaluating self-management interventions: Losing focus on what we wish to achieve

Outcome measures or themes varied greatly between studies and even within SMI aiming at a specific diagnostic group, as also reported by others [19]. Health outcomes predominated, which is not surprising given the focus on medical management. Remarkably, however, some studies that focused on a (partially) medical management intervention did not measure health outcomes. Likewise, some medical management interventions were evaluated with psychological outcomes, and an emotion management intervention was evaluated on knowledge of the disease. It seems that current evaluation studies tend to lose focus on what interventions are aimed at, which also

hampers conclusions about their effectiveness. Others have recognized this, too, and recommend use of a core set of measurement outcomes to evaluate SMI [19,22,123].

A content-based framework for the selection of outcome measures or groups of outcome measures

The framework presented in Figure 3 proposes a start for a more standardized evaluation approach for SMI for young people with chronic conditions. The outcomes matched those in comparable reviews [18,19], which strengthens the validity of the framework. It may be used to select outcome measures on the basis of the specific content of interventions (as described and numbered per domain in Table 3). However, the classification is broad and measures must be selected based on the goal of the intervention and the measurement properties of the measure. Further sharpening requires more studies into outcomes and measurement instruments.

A fact worth mentioning is the lack of qualitative evaluation studies for SMI. Since qualitative research delves into the contexts of interventions, we recommend future studies to employ a mixed-methods design including a qualitative component. This would help identify ‘effective ingredients’ of SMI and answer the question of what works for whom [124]. The outcome measures in our framework may serve as themes for qualitative research, but themes related to the characteristics of interventions need to be included as well.

Strengths, limitations and other considerations

This study included a systematic and comprehensive search, and was the first to review content of pediatric SMI and classify interventions using a broad self-management framework. Other recent reviews in this field that focus particularly on children and/or adolescents (0-18 years), aimed at researching the effectiveness of SMI and included only RCT’s or studies with repeated measures designs [18,19]. In contrast, our study shed light on the broad content and range of today’s self-management support for young people with chronic conditions. As such, we dealt with the more fundamental question of what exactly is meant by self-management and self-management support. Furthermore, by matching content of SMI and outcome measures used, a selection tool for future evaluation studies was presented. This also corresponds to the fundamental question of what might be expected from self-management support, and provides a first step towards a much-needed general evaluation framework for different types of interventions.

Lorig & Holman’s model is often referred to in the self-management literature and seems valid to classify SMI in children, adolescents and young adults, because our results

showed that SMI aimed at certain domains of self-management are not exclusive for age groups. This does not imply that certain content is applicable to all ages; for example, vocational participation is more relevant for older adolescents than for younger children. Differences between age groups should therefore be taken into account when evaluating SMI.

This study looked at many types of SMI across a range of chronic conditions. This may be a limitation, because our search terms did not include specific chronic conditions and we might have missed studies that did not include specific key words from our search. However, we feel this is always an issue when performing a systematic literature review which probably is more related to the way databases are organized than to the sensitivity of our search strategy. Furthermore, our non-categorical approach may also be a strength, because it enables a more general view on self-management irrespective of diagnosis. This is relevant because these young people face comparable challenges and similar adaptive tasks irrespective of type of condition [4,115]. Yet, they may need different support in view of individual socio-demographic and psychological factors [117]. In this respect young people within a specific diagnostic group may differ as much as those in different diagnostic groups [125]. Interestingly, only 7% of the SMI found in the present study were developed for chronic conditions in general. Since specific paediatric diagnostic groups are often small, achieving effectiveness and cost-effectiveness of disease-specific SMI would be problematic [20]. A more generic approach with a disease-specific component for different diagnostic groups may be more convenient [4], and should not be problematic since the core elements of self-management support are the same across different approaches [126]. An example is the 'Skills for Growing Up' tool developed in paediatric rehabilitation and adjusted on disease-specific content for use in paediatric nephrology [127].

Gaining insight into effectiveness of different types of interventions was hindered by the heterogeneity in outcome measures. Most studies in this review were from Western countries, and interventions for young people with diabetes or asthma predominated. These conditions generally include a burdensome medical regimen, which may have added to the focus on medical management. Yet, a sub-analysis (not presented in this paper) showed that even after removing diabetes and asthma studies, the focus still remained on medical management alone than on other self-management domains.

CONCLUSIONS

The content of different SMI relate to self-management tasks of people with chronic conditions, and self-management skills they should develop. Yet, healthcare professionals should be aware of the importance of role and emotion management in self-management. Also, in view of these young people's developmental challenges, an experiential approach focusing on learning (from others) and 'mastery experiences' might be more appropriate in pediatric care. Future evaluations should provide details about theoretical bases of interventions, and should match evaluation outcomes and themes to intervention content and characteristics. The content-based evaluation framework presented in this study may assist in this, while further research might help identify valid outcome measurement instruments. Mixed-methods research is recommended to gain more insights in the contexts, including social context, and working mechanisms of SMI.

PRACTICE IMPLICATIONS

Self-management support is important for people with chronic conditions to help them deal with their condition in daily life. This is even more pertinent to young people growing up with chronic conditions, who have to face the normal tasks of development (e.g., acquiring autonomy) and have to engage in lifelong medical management of their condition. Therefore, it remains important to research the effects of SMI. Future evaluation studies should make sure that their evaluation outcomes match with the content and characteristics of the SMI, and may benefit from the use of more generic outcome measures in SMI evaluation. Our content-based evaluation framework and overview of SMI content, characteristics and outcomes may assist researchers in doing so. Furthermore, our overview may give clinicians and other healthcare professionals insight into the broad range of self-management and self-management support, and as such may assist them in determining the breadth and focus of the support they provide.

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Appendix A. Overview of study and intervention characteristics (n=86)*

Study characteristics			Participants		Intervention characteristics		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists
Barakat et al. 2010	United States of America	RCT	Sickle cell disease	14.1	Individual	Home environment	Doctoral students in clinical psychology or psychologists
Barrera & Schulte 2009	Canada	Cohort	Cancer	12.7	Group	Clinic (Pediatric center)	Psychologist, and clinical assistants
Beebe et al. 2010	United States of America	RCT	Astma	7-14	Group	School	NA
Bekesi et al. 2011	Hungary	Cohort	Oncology, diabetes, juvenile immune arthritis	13.3	Group	Camp, and clinic (medical centre)	Volunteers (selected and trained, profession not mentioned)
Berrien et al. 2004	United States of America	RCT	HIV	10 .0	Individual home visit	Home	Experienced registered nurse
Betz et al. 2011	United States of America	RCT	Spina Bifida	16.0	Group	Clinic	Trainer (not mentioned if this was a healthcare professional)
Bultas et al. 2013	United States of America	Cohort	Heart disease	8-15	Group	Camp	Pediatric nurses and cardiologists
Burkhart et al. 2007	United States of America	RCT	Asthma	7-12	Group sessions	Clinic (university center for nursing reserach)	Research associate
Butz et al. 2005	United States of America	RCT	Asthma	8.0	Group sessions	School	Asthma educator (not mentioned if this was a healthcare professional)
Cafazzo et al. 2012	Canada	Cohort	Diabetes	14.9	Individual	Online	Clinicians
Canada et al. 2007	United States of America	Cohort	Cancer	21.3	Individual	Clinic	Doctoral level clinical psychologist

Self-management interventions for young people with chronic conditions

Intervention characteristics					
Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base	
Educational sessions	Daily paper-and-pencil pain diaries, homework, review of homework, and biweekly check-in telephone calls	Sessions take 90 minutes. Intervention: 4 sessions (3 sessions, 2 weeks apart with a booster session 1 month later)	MM	NA**	
Skills training sessions	Fun activities and games guided by cognitive behavior strategies and expressive therapies such as music, art, and drama.	A 2-hr group session once a week for 8 weeks.	RM and EM	NA	
Art therapy sessions	Art making, and sharing feelings related to the art created.	A 1- hour session for 7 weeks.	MM and EM	NA	
Camping program	Adventure-based program with activities and fun.	NA	RM	NA	
Educational sessions	A comic book, a video, filling out notebooks with rewards (prizes) if they did, and eventually role reversal.	NA	MM	NA	
Skills training sessions	Making a transition plan, and practicing strategies to obtain goals, i.e. role playing, one-to-one consultation, coaching, reinforced learning, use of audio visual aids, accessing the Internet, and mentored learning.	2-day workshop	MM and RM	NA	
NA	NA	5 days and 4 nights	NA	NA	
Educational sessions	Educational videos, and homework assignments	NA	MM	(Cognitive) social learning theory	
Skills training sessions	Practiced and demonstrated specific skills (peak flow meter and inhaler technique). Discussions with the educator, and a coloring book, a peak flow meter, and a spacer device were given to children.	4 hours of instruction during two separate sessions	MM	NA	
Telemanagement system	Reminders, cueing, social media communication, and the gamification of routine management tasks.	NA	MM	NA	
Educational (and support) sessions	Workbook, homework, and follow-up phone calls	NA	RM and EM	NA	

Appendix A. Overview of study and intervention characteristics (n=86)* (*continued*)

Study characteristics			Participants		Intervention characteristics		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists
Channon et al. 2007	United Kingdom	RCT	Diabetes	15.3	Individual	Home	Nurses, and a health psychologist
Chiang et al. 2009	Taiwan	RCT	Astma	6–14	Individual	Home, and clinic	Nursing graduate student
Christian & D'Auria 2006	United States of America	RCT	Cystic Fibrosis	8–12	Individual home visit, and group session	Home, and clinic	NA
Clark et al. 2004	United States of America	RCT	Astma	7–10	Goup	School	NA
Clark et al. 2010	United States of America	RCT	Asthma	11.9	Group lessons	School	Graduate students, and community leaders trained in the program (profession not mentioned)
Connelly et al. 2006	United States of America	RCT	Migraine	7–12	Individual	Home	NA
Creedy et al. 2004	Australia	Cohort	Various chronic conditions	10–14	Group sessions	NA	Graduates of a leadership training course co-facilitated the intervention with healthcare professionals
Curle et al. 2005	United States of America	Qualitative	Various chronic conditions	7–12	Group sessions	Clinic (specialized unit)	A clinical psychologist, occupational therapist, mental health nurses, and specialist pediatric nurses or social workers.

Self-management interventions for young people with chronic conditions

Intervention characteristics					
Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base	
Motivational interviewing sessions	Awareness building, problem solving, and goal-setting.	The frequency and location of appointments was determined by the participants to fit with the patient-driven principles of motivational interviewing. Interviews lasts between 20 and 60 min.	NA	NA	
Skills training sessions	Relaxation training	During the 12-week intervention, participants practiced relaxation for 30 min at least three times per week.	MM	NA	
Educational sessions and support sessions	A computer software program, a notebook with worksheets, and a journal (individual). Discussion and peer contact	NA	RM	NA	
Educational sessions	Handouts and homework assignments, group discussions, asthma action plan.	NA	MM	NA	
School program	Interactive problem-solving activities. Peer education component (not necessarily Asthma patient) in which peers educated asthma awareness to participants through skits, creative dramas or music.	NA	MM, RM and EM	NA	
CD-ROM	Educational modules, active coping plan, and gamification (with feedback).	NA	MM and EM	NA	
Family sessions (parallel but seperate sessions for parents and children).	Peer support	8 weeks	RM and EM	NA	
Family sessions (parallel but seperate sessions for parents and children).	Play therapy, narrative therapy, relaxation training and group work.	6-8 sessions	EM	Cognitive behavioral theory, and Systemic theory	

Appendix A. Overview of study and intervention characteristics (n=86)* *(continued)*

Study characteristics			Participants		Intervention characteristics		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists
Cushner-Weinstein et al. 2007	United States of America	Cohort	Epilepsy	7-17	Group sessions	Camp	Medical professionals, and counseling staff
Davis et al. 2004	United States of America	RCT	Cystic Fibrosis	7-17	Individual	Home	NA
Dobson 2014	United States of America	Cohort	Sickle cell disease	6-8	Individual	Clinic and home	Child life specialist
Downs et al. 2006	Australia	RCT	Cystic Fibrosis	8.4	Individual	Clinic	Caregivers
Dufresne et al. 2013	France	Cohort	Ichthyosis	6 and older	Group	Reception centre	Physician and paramedic team member
Eccleston et al. 2003	United Kingdom	Cohort	Chronic pain	14.3	Individual and family-centered	Clinic	A paediatric rheumatologist, clinical psychologist, physiotherapist, occupational therapist, and a nurse

Self-management interventions for young people with chronic conditions

Intervention characteristics					
Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base of SM	
Camping program	Traditional camp activities (rope course, swimming, arts, crafts etc.) combined with activities with condition-specific goals and relevance, and support groups (peer contacts).	7 days	MM, RM and EM	NA	
CD-ROM	Educational modules	NA	MM, RM and EM	NA	
Diary and guided imagery, including one training session	Participants kept a diary, recording their daily activities and all pain episodes, including location and intensity, as well as strategies for management. The diaries included blank daily pages with the instruction, "Use one page a day to describe your activities and your pain, and one page to draw a picture".	The training sessions lasted from 15-45 minutes	MM	Cognitive behavioral therapy	
Cognitive behavioral therapy sessions	NA	10-week period, with each of the 10 chapters taking approximately 20 minutes to complete	MM	(Cognitive) social learning theory	
Two sessions called 123 Tem'peau sessions And a game with a set of multiple-choice questions was used.	Children and siblings: "What is ichthyosis? Why do I need the cream? Why am I sick? Is it normal to have pain? What about school and me? What about the hospital and me?" Parents, children and siblings >12 years: "What is ichthyosis? What are the treatments? What is genetics? What is functional management? What are my social rights?" The game addressed various topics: therapy, genetics, care, pain, rehabilitation and social rights.	Two sessions of two hours each	MM+RM	NA	
Educational sessions, and cognitive behavioral therapy sessions.	Many sessions required evening or weekend written and skills practice homework. Patients received written information about all aspects of the programme which built into a patient manual.	Overall contact time was 110 hours (60 hours of physical and occupational activity; 35 hours of cognitive therapy, and 15 hours education). Each session lasted 50 minutes. The day was structured as a school day from 9 00 am to 3 45 pm.	MM and EM	(Cognitive) social learning theory, and Cognitive behavioral theory	

Appendix A. Overview of study and intervention characteristics (n=86)* *(continued)*

Study characteristics			Participants		Intervention characteristics		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists
Franklin et al. 2006	United Kingdom	RCT	Diabetes	11-16	Individualized	Clinic, and by phone	Diabetes healthcare team
Fuchs et al. 2013	Netherlands	Cohort	Juvenile idiopathic Arthritis	14-19	Individual	NA	Child psychologist and counselor/philosopher
Gerber et al. 2007	United States of America	Cohort	Diabetes	22.3 yrs	Individualized, but also online discussion with peers possible	Online	Psychologist, patient advocacy expert and social worker
Grey et al. 2009	United States of America	RCT	Diabetes	9.9 yrs	Group sessions	Clinic	Mental health professional
Hackett et al. 2005	United Kingdom	Cross-sectional	Juvenile Idiopathic Arthritis	Median: 14 yrs	Group	Camp	Occupational therapist, Physiotherapist, Nurse / therapist, assistant
Hampel et al. 2003	Germany	Cohort	Astma	11.6 yrs	Group	Clinic (inpatient)	Psychological, educational, and medical staff
Hanauer et al. 2009	United States of America	RCT	Diabetes	17.9 yrs	Individualized	Online	NA
Hayutin et al. 2009	United States of America	Cohort	Inflammatory Bowel Disease	13-17 yrs	Group sessions	NA	NA

Self-management interventions for young people with chronic conditions

Intervention characteristics				
Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base
Telemedicine system (text-messages on phone)	Individual goal-setting at clinic. Automated delivery of a series of messaging, including a weekly reminder of the goal set, and a daily message providing tips, information or reminders to reinforce this goal (by phone).	NA	MM	(Cognitive) social learning theory
Narrative self-reflections	Phase 1: self-investigation, about important life experiences; Phase 2: process-promoting, about daily situations and coping, Phase 3: second self-investigation, consistencies and changes in person narratives;	Phase 1 included one SMC session. Phase 2 consists of 6 weekly individual sessions of about 1 h each. Phase 3 consists of 3 weekly individual sessions of about 1 h each.	RM+EM	Self-confrontation method
Telemedicine system (web-based)	Educational module, and goal-setting exercises with individualized feedback, role-playing, group discussion, empowerment activities, and communication skills training to improve interactions with health professionals. There was a discussion board available, and there were three 'ask the experts segments'.	NA	MM	NA
Educational sessions	Role play, coaching, and practice at home	6 weekly sessions	RM and EM	(Cognitive) social learning theory
Camping program	Climbing, canoeing, abseiling and a trip to a theme park.	4 day annual event	RM	NA
Family sessions (separate for parents and children)	Educational group work	4 weeks stay, 10 1 h-long training sessions.	MM and EM	Stress theory
Telemedicine system (web-based)	BG diaries (blood glucose check) and two daily factoids of which one was related to diabetes and one related to unusual fun facts or trivia. System sends reminders to check BG, and gives (positive) feedback.	NA	MM	NA
Family sessions (separate for parents and children, except for relaxation session and conflict resolution communication session)	Review of written homework, didactic presentations, discussion, problem solving and practice of the new skill, plans for application during the week, and assignment of homework related to the skills.	10 sessions	MM	Cognitive behavioral theory

Appendix A. Overview of study and intervention characteristics (n=86)* *(continued)*

Study characteristics			Participants		Intervention characteristics		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists
Hechler et al. 2010	Germany	Cohort	Chronic pain	14.0 yrs	Individualized	Clinic (inpatient)	Therapists
Herbert et al. 2013	United States of America	Qualitative	Diabetes (type 1)	11-14	Group	Clinic	Study team counselors, but not mentioned who these were
Hilberink et al. 2013	Netherlands	Mixed methods	Cerebral palsy, spina bifida, Neuromuscular disease	19.9 yrs	Group	School, and rehabilitation clinics	Pedagogues, psychologists, social workers, a sexologist, and a teacher
Højberg et al. 2010	Denmark	Cohort	Congenital physical disability	18-25 yrs	Group sessions	Rehabilitation clinic (and a trip to Lithuania)	Occupational therapist, and a socio-educational assistant (if necessary another assistant was hired)
Huss et al. 2003	United States of America	RCT	Asthma	9.6 yrs	Individual	Home	NA
Jan et al. 2007	Taiwan	RCT	Asthma	10.5 yrs	Individual	Clinic (outpatient)	NA
Jones et al. 2010	United States of America	RCT	Cancer	12-18 yrs	Individual	Home	Trained healthcare professionals
Joseph et al. 2007	United States of America	RCT	Asthma	15.3 yrs	Individual	School	NA
Kashikar-Zuck et al. 2012; 2013	United States of America	RCT	Juvenile Fibromyalgia	15.0 yrs	Individual	Clinic	Therapists with postdoctoral training in pediatric psychology

Self-management interventions for young people with chronic conditions

Intervention characteristics					
Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base	
Skills training sessions	Interceptive exposure; bilateral stimulation in the form of tapping; and cognitive coping strategies to reduce pain intensity.	NA	MM	NA	
TeamWork. Adolescent-parent type 1 diabetes (T1D) program developed to prevent deterioration in diabetes care among adolescents with T1D. Coping skills or education group with a study team counselor at four consecutive regularly scheduled diabetes clinic visits.	Coping skills session topics included communication and diabetes management, problem solving to improve blood glucose management, healthy food choices and avoiding arguments, and how attitudes affect behaviors and how these relate to physical activity. Each session started with an overview of the skill and was followed by a discussion about what was typical for the family and how they could use the skill in daily life.	NA	MM	NA	
Educational/support sessions	Homework assignments	7 sessions (90 min each, scheduled over a 12-week period).	RM and EM	Flirt model	
Developmental instructional training course (skills training sessions)	NA	The group met 20 hours per week, 4 days a week. Approximately every sixth week out of house activities of 2-3 days duration. In addition: a 10-day study trip to Lithuania.	RM	NA	
CD-ROM	Interactive game including levels and quizzes and feedback from an on-screen nurse.	NA	MM	NA	
Telemedicine system (Internet-based)	Educational modules, electronic diary, action plan, and monitoring system	NA	MM	NA	
CD-ROM	Information videos, text, stories, and a game.	NA	MM and RM	NA	
Telemedicine system (web-based)	Theory-based health messages and information on Asthma control. Normative (compared with other students) and ipsative (compared with your last session) feedback.	NA	MM	Transtheoretical, and Health belief model	
Cognitive-behavioral therapy sessions (in 3 of the 8 parents were also present)	Education and skills training. Instructions for home practice were also reviewed with participants.	8 weekly individual sessions, and 2 booster sessions.	MM	Cognitive behavioral theory	

Appendix A. Overview of study and intervention characteristics (n=86)* *(continued)*

Study characteristics			Participants		Intervention characteristics		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists
Kato et al. 2008	United States of America	RCT	Cancer	13-29 yrs	Individual	NA	NA
Koontz et al. 2004	United States of America	RCT	Sickle cell disease (anemia)	8-12 yrs	Group session	School	Teachers
Kouba et al. 2013	United States of America	Cohort	Asthma	15.9	Group and individual	School	Registered nurses, dietitian and dietetic interns
Krishna et al. 2003	United States of America	RCT	Asthma	7-17 yrs old	Individual (clinical staff leaves when the patient is working on IMPACT)	Clinic	Multidisciplinary team involved in determining content of the program.
Kumar et al. 2004	United States of America	RCT	Diabetes	13.6 yrs	Individual	Online	NA
Laffel et al. 2003	United States of America	RCT	Diabetes	12.1 yrs	Individualized, child with parent (during office visits)	Clinic	Research assistant

Self-management interventions for young people with chronic conditions

Intervention characteristics					
Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base	
CD-ROM (PC-game)	Game	NA	MM	Self-regulation theory, and (Cognitive) social learning theory	
School program	Peer education	1 hour	MM and RM	NA	
The ICAN program is thus composed of four elements: (1) asthma education, (2) nutrition education synthesized with CST, targeting obesity prevention and management, (3) reinforcement visits with a registered nurse (RN) and dietetic intern, and (4) a family information meeting. Sessions included group problem-solving and role-play activities, which allowed opportunities to increase perceived competence and foster peer support for improved health choices. In addition electronic modules were developed and offered.	Quest for the Code, an interactive, three-dimensional video that combines asthma education with rich graphics and top celebrities, was viewed by the students in groups during lunch periods. The second session included a review, discussion about the group's asthma symptoms, medications, management, and additional content from the "Fight Asthma Now" program developed by the Respiratory Health Association. Each nutrition session included a focused nutrition lesson followed by introduction of a coping skill. Then interactive exercises were completed with students allowing them the opportunity to practice the coping skill with a nutrition scenario. The CST strategies were used to reinforce nutrition information. CST is a cognitive behavioral strategy that teaches students personal and social coping skills to assist in making health-related decisions. Electronic modules: These interactive modules encouraged students to seek resources and make decisions related to the target behaviors using existing Internet resources.	The ICAN educational session covered an 8-week period including the makeup session. This was followed by two reinforcement visits. The total program length spanned 14 weeks.	MM	Orem's Self-Care Deficit theory of Nursing	
Telemedicine system (interactive multimedia program)	Animated lessons, each averaging a minute in length. Each template was designed to illustrate concepts, test comprehension and reinforce learning, develop decision-making skills or improve a child's ability to communicate their asthma.	During office visits	MM	NA	
Telemedicine system (personal device)	Game with rewards	NA	MM	NA	
Family sessions (for child and parent together)	Written materials, discussion, making a responsibility-sharing plan	During office visits	MM	NA	

Appendix A. Overview of study and intervention characteristics (n=86)* *(continued)*

Study characteristics			Participants		Intervention characteristics		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists
Løding et al. 2008	Norway	Cohort	Diabetes	13-17 yrs	Group sessions	Clinic (outpatient)	A team of nurses, medical doctors, and a trained social worker
MacDonald & Greggans 2010	United Kingdom	Qualitative	Cystic Fibrosis	8-18 yrs	Individual	Home/public environment	Volunteers (after following a training programme)
Martin et al. 2009	United Kingdom	Cohort	Diabetes	9-11 yrs	Group sessions	Clinic	Pediatric diabetes nurse specialist, assistant psychologist under supervision of a consultant (pediatric clinical psychologist).
Maurice-Stam et al. 2009	Netherlands	Cohort	Cancer	8-12 years	Group sessions	Clinic (outpatient)	Dermatologists or pediatricians, psychologists, and dietitians
McClellan et al. 2009	United States of America	Cross-sectional	Sickle Cell Disease	8-20 yrs	Group session and individual device	Clinic, and by phone	NA
McDonagh et al. 2007	United Kingdom	Cohort	Juvenile Idiopathic Arthritis	14.2 yrs	Individual	Clinic	Project coordinator from clinic assisted by local consultant rheumatologist
Meade et al. 2003	United States of America	Cross-sectional	End-stage renal disease	13.7 yrs	Group sessions	Clinic	A nurse from the transplant team, and a nephrologist.
Merlijn et al. 2005	Netherlands	Cohort	Pain	14-18 yrs	Group sessions, and individual telephone contacts	Clinic	NA

Self-management interventions for young people with chronic conditions

Intervention characteristics					
Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base	
Separate support/ educational/skills sessions for adolescents and parents	Warm-up activity such as painting, movement/exercises, or a round of questions about topics that were not related to diabetes, followed by group discussion.	NA	MM, RM and EM	NA	
Peer-support	Mentorship	NA	EM	NA	
Educational sessions	Educational stories about diabetes, group discussions, coloring/drawing pictures, one-to-one computer session.	NA	MM and EM	(Cognitive) social learning theory	
Educational/skills/support sessions	Modelling, contingency management, exposure exercises and cognitive techniques.	NA	MM, RM and EM	Cognitive behavioral theory	
Skills training session, and telemedicine system (personal device)	Daily pain diary and coping skills practice (through audio files).	NA	MM	Cognitive behavioral theory	
Individual (transition) plan	Age and developmentally appropriate information resources, and goal-setting	NA	MM and RM	NA	
Educational/skills/support sessions	Problem solving, stress management/ relaxation, talking with other teens and/or parents, communication role-plays, questions and answers with the nephrologist, medication presentation by the nurse.	2 4-hour sessions	MM	NA	
Educational and skills training sessions (two sessions were for parents only: one at the beginning and one at the end). The others were for adolescents only.	Training and exercise book, peers, written material, exercises and homework assignments.	NA	MM, RM and EM	(Cognitive) social learning theory	

Appendix A. Overview of study and intervention characteristics (n=86)* *(continued)*

Study characteristics			Participants		Intervention characteristics		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists
Nansel et al. 2007	United States of America	Cohort	Diabetes	11-16 yrs	Individualized	Home or public environment	Trained non-professionals (bachelor degree and/or graduate students in health related fields)
Newcombe et al. 2012	Australia	RCT	Chronic respiratory condition	13.4 yrs	Individualized, but also online conversation with peers	Online	NA
Newton & Ashley 2013	United States of America	RCT	Diabetes (type 1)	13-18	Individual, but online contact with peers	Online	Moderators, but not mentioned who these were
Ng et al. 2008	China	RCT	Asthma	9.2 yrs	Group sessions	Clinic	NA
O'Mahar et al. 2010	United States of America	Cohort	Spina bifida	16.6 yrs	Group and individual	Camp	Counselors, but not mentioned who these are.
Holbein et al. 2013							
Palermo et al. 2009	United States of America	RCT	Chronic pain	14.8 yrs	Individualized	Online	Online therapist: a psychology postdoctoral fellow who had one year of specific experience in delivery of face-to-face CBT
Fales et al. 2014							

Self-management interventions for young people with chronic conditions

Intervention characteristics					
Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base	
Skills training sessions with motivational interviewing incorporated, supplemented with telephone calls.	Reviewing self-monitoring records, goal-setting, going through the steps of behavior change, brainstorming about possible solutions of difficulties encountered (problem-solving). The personal trainers provided suggestions, encouragement, and positive feedback.	6 sessions	MM	NA	
Telemedicine system (educational website)	Information on web-site, daily diary, assignments/homework and peer contact.	NA	MM	NA	
Telemedicine system: interactive web-based intervention with problem solving through discussion in forums, chat rooms and blogs	Three types of asynchronous discussions were held in the forums: 1) General discussion on the weekly topic. 2) Diabetes-related scenarios discussing psychosocial diabetes-related issues. 3) Open discussion where participants could post their own problems to the group. Weekly topics were: Frustrations with diabetes, Benefits of good control, Family, Friends, Body image, exercise and diet, Community, School and sports, Worries about diabetes.	7-week, weekly topics	MM+RM+EM	Bandura's Self-Efficacy Theory	
Educational/support sessions	Joint activity (to talk about take-home tasks from previous meeting), parallel groups (children and parents work separately on a common theme), joint activity (discussion).	5 educational sessions, and 6 support sessions.	MM, RM and EM	NA	
Camping program with support sessions	NA	11 2-hour group sessions	RM and EM	NA	
Telemedicine system (web-based program).	Treatment modules with video files and audio files, and daily diary. Also queries (assignments).	NA	MM	(Cognitive) social learning theory, and Cognitive behavioral theory	

Appendix A. Overview of study and intervention characteristics (n=86)* *(continued)*

Study characteristics			Participants		Intervention characteristics		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists
Payne et al. 2013	United States of America	Case study	End-stage renal disease	14-16	Individualized	Clinic	NA
Pulgaron et al. 2010	United States of America	RCT	Asthma	10.4 yrs	Group sessions	Camp	Interventionists were clinical psychology graduate students, trained and supervised by two pediatric psychologists and an advanced graduate student; interventionists were not camp counselors.
Raghavendra et al. 2013	Australia	Mixed methods	CP, physical disability or acquired brain injury	10-18	Individualized	Home	Speech pathologist and research assistants
Rami et al. 2006	Austria	RCT	Diabetes	10-19 yrs	Individualized	Online/Phone	NA

Self-management interventions for young people with chronic conditions

Intervention characteristics					
Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base	
Six modules: four individual and two joint (with parents). A quiz was given after each session to assess knowledge of skills before progressing to the next module.	Sessions were designed to identify and address knowledge, behavioral, and cognitive barriers to medication adherence. Session 1: knowledge and education about regimen, 2) sharing regimen responsibilities (joint session), 3) review of problem-solving skills and solutions (joint session), 4) role of cognitions as adherence barriers, 5) emotional regulation and coping skills to overcome barriers to adherence, 6) review of skills learned throughout intervention.	The intervention was completed in a minimum of 6 weeks. Including data collection, sessions lasted 60 to 90 minutes.	MM	Health Belief model	
Camping program with skills training sessions	Group discussion, modeling of how to apply the PAC-T, and role plays. Participants received personal manuals with blank fill-in sheets to list potential solutions to each problem.	NA	MM	NA	
Educational and skills training sessions	Focused on: identifying and evaluating the family's current computer and Internet access; teaching the youth and family about cyber safety including the use of filtering and privacy settings, the development of house rules for expected online behaviour, and explanations/ discussion of age-appropriate social media; providing appropriate software and equipment based on the goals; providing training materials including visual supports or instructions as well as hands on training and practice to use the software, equipment or Internet; providing interface solutions, such as speech recognition software for text entry, word-prediction software or screen reading software. Training was framed to ensure youth accessed the identified web sites to engage in social networking with their peers, friends and families.	NA	RM	NA	
Telemedicine system (on phone)	Monitoring through daily diaries.	NA	MM	NA	

Appendix A. Overview of study and intervention characteristics (n=86)* *(continued)*

Study characteristics			Participants		Intervention characteristics		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists
Rhee et al. 2011	United States of America	RCT	Asthma	14.6 yrs	Group sessions	Camp	Peer leaders selected by school teachers/ nurses or clinicians
Rhee et al. 2012	United States of America	RCT	Asthma	17.6 yrs	Training sessions	Camp	Peer leaders selected by school teachers/ nurses or clinicians
Sattoe et al. 2013	Netherlands	Mixed methods	End-stage renal disease	16-25	Group	Camp	Pediatric nephrology health care professionals
Shames et al. 2004	United States of America	RCT	Asthma	8.0 yrs	Individualized	Clinic, and home environment	Case manager, allergist / immunologist, and pediatric nurses
Smith Fawzi et al. 2012	Haiti	Mixed methods	HIV	10-17 yrs	Group sessions	Clinic	Social workers
Staab et al. 2006	Germany	RCT	Atopic dermatitis (Eczema)	8-18 yrs	Group sessions	Clinic	Dermatologists, pediatricians, psychologists, and dieticians
Stinson et al. 2010	Canada	RCT	Juvenile Idiopathic Arthritis	15.6 yrs	Individualized, but contact with peers possible	Online	Trained coach: non healthcare professional with an undergraduate degree in psychology
Stulemeijer et al. 2005	Netherlands	RCT	Chronic Fatigue Syndrome	15.5 yrs	Individualized	Clinic	Child therapists who were trained and supervised by an experienced cognitive behavioral therapist

Self-management interventions for young people with chronic conditions

Intervention characteristics					
Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base	
Camping program	Peer leaders led group activities that involved discussion, strategic thinking, knowledge-testing games, and role playing.	Each session lasted approximately 45-60 minutes.	MM and RM	NA	
Camping program	Disease-specific activities (sessions)	NA	NA	NA	
Camping program: with different elements: workshop present yourself, movie making workshop, dancing workshop, sports, cooking teams, free time, theater performance by professional artists, art workshop, drumming workshop, acting workshop. Also: buddy-attendant concept (peer-to-peer support).	Aimed at independent living with ESRD and developing self-management skills, but no focus on disease-specific issues; about transition to adulthood in general, i.e. aimed at general competencies like self-confidence, self-efficacy etc.	Five days	RM+EM	NA	
Educational and skills training sessions	Informational books, phone calls by interventionists, video game, asthma management plan	NA	MM	NA	
Family sessions (child-parent pairs)	Sharing experiences	The sessions were implemented over a one year period (held bi-monthly).	MM, RM and EM	(Cognitive) social learning theory	
Educational sessions and skills training	Sharing experiences, practicing skills	NA	MM and EM	NA	
Telemedicine system (web-based)	Goal-setting (diary), homework, knowledge quizzes, discussion board, feedback from coach	NA	MM, RM and EM	NA	
Cognitive-behavioral therapy sessions	NA	10 sessions	MM, RM and EM	Cognitive behavioral theory	

Appendix A. Overview of study and intervention characteristics (n=86)* *(continued)*

Study characteristics			Participants		Intervention characteristics		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists
Ten Hoedt et al. 2011	Netherlands	RCT	Phenylketonuria	15 yrs	Individualized	Online	Dietician
Torok et al. 2006 ⁶⁵	Hungary	Cohort	Diabetes, oncologie	Oncologie 15.58 yrs; Diabetes 14.90 yrs	Group	Camp	NA
Velsor-Friedrich et al. 2005	United States of America	RCT	Asthma	10.1 yrs	Group sessions, and inividualized nurse visit	School	Nurse practitioner
Verhoef et al. 2014	Netherlands	Cohort	Physical disability	16-25	Group and individual	Clinic (outpatient)	Occupational therapist, psychologist, jobcoach, social worker
Wang et al. 2008	United States of America	Cohort	Diabetes	14.0 yrs	Group sessions	Camp	Physician, medical students, and a dietitian
Wysocki et al. 2007	United States of America	RCT	Diabetes	14.0 yrs	Family-centered	Clinic	Psychologists

Self-management interventions for young people with chronic conditions

Intervention characteristics					
Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base	
Telemedicine system (secured website)	Monitoring through daily diaries	NA	MM	NA	
Camping program	Disease-specific activities and reinforcement of positive thinking.	2 separate 8-day sessions	NA	NA	
Educational sessions	Interactive teaching approach utilizing group discussion, stories, games, and role-play.	Six weekly sessions	MM and RM	Self-care theory	
Educational and skills training sessions	Group sessions provided information and discussion about work-related topics and aimed to increase insight into personal (dis)abilities, addressing work objectives, coping strategies (dis)abilities, addressing work objectives, coping strategies, occupational balance, finding a (suitable) job, how to present oneself at a job interview, and employment regulations and social security. In addition, group sessions offered opportunities to develop communication and interaction skills, to share experiences, and to increase group members' self-efficacy. Homework assignments, for example preparing a self-presentation or preparation for a job interview, contributed to active participation and planning. Individual sessions: developing vocational skills and work routines, enhancing self-management skills, providing work placement opportunities and work experience, and advising on workplace modifications.	Group support programme consisting of six 2-hour sessions over 8 weeks.	RM and EM	Model of human occupation (MOHO)	
Camping program	Educational sessions	NA	MM	NA	
Educational and skills training sessions	Problem solving training, communication training, and cognitive restructuring and functional-structural family therapy. Therapists participated actively, frequently providing instructions, feedback, modeling, and rehearsal. Also homework was given each session.	12 sessions over 6 months attended	MM and RM	NA	

Appendix A. Overview of study and intervention characteristics (n=86)* *(continued)*

Study characteristics			Participants		Intervention characteristics		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists
Xenakis et al. 2010	United States of America	Cohort	Physical disabilities, 93% congenital	14-21 yrs	Individualized and group sessions	Clinic / Hospital based center	A program coordinator, instructors and tutors, a care assistant, and volunteers. The instructors, experts in their chosen fields, have prior experience working with adolescents and/or persons with physical disabilities
Yoon et al. 2007	United States of America	Cohort	Sickle Cell Disease	10.7 yrs	Individualized	Clinic	NA

*Abbreviations: NA=Not available (meaning this information couldn't be derived from the original research article); RCT=Randomized Controlled Trial; SM=Self-management; MM=Medical management; RM=Role management; EM=Emotion management

Self-management interventions for young people with chronic conditions

Intervention characteristics				
Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base
Educational sessions	Discussions, expressive arts, goal-setting, field trips (community recreation and learning institutions).	12 consecutive 2½-hour weekly sessions	MM, RM and EM	NA
CD-ROM	Game with feedback	NA	MM	Gaming theory

Appendix B. Overview of outcome measures used in the evaluation studies*

Groups of outcomes	Specific outcomes	References
Health outcomes	Clinical outcomes (e.g. blood glucose levels, peak expiratory flow rate or pulmonary functioning, BMI, fatigue)	[27, 30, 38, 39, 41, 44-47, 51-53, 55-57, 64, 66, 99, 101]
	Number of (outpatient) clinic visits or disease-related emergency department visits	[35, 39, 44, 64, 105]
	Hospitalizations (due to chronic condition)	[35]
	Frequency or intensity of disease-specific symptoms, i.e. morbidity (e.g. asthma episodes or pain)	[28-33, 35, 36, 38, 39, 41-45, 66, 73-77, 83, 91, 93, 96, 100, 105, 109]
	Activity limitations or functional or disease-related disability / perceived difficulty in performing daily activities	[73, 75, 77, 91, 93-95]
	Medication or treatment adherence or usage	[31, 34, 35, 38, 41, 44, 46, 55-57, 64, 79, 85, 90, 96, 101]
	Self-perceived health status	[41]
	Somatic awareness	[74]
	Disease-specific coping (e.g. coping with pain during SCD-related pain episodes)	[95, 105]
	Functional health status / Physiologic health status / Physical functioning or activity	[64, 66, 74, 81, 94, 99]
	Condition-related disability	[100]
	Goal status in individual health and wellness planning	[103]
	General satisfaction with health	[40]
	Condition-specific goals	[11]
	Risk behavior (e.g. smoking)	[35]
Self-care	Self-care behavior or practice	[39, 44, 46, 110]
	Disease-related skills	[65]
	Independent health behavior (e.g. self-medication or independent consultations)	[98]
	Motivation, knowledge, skills and abilities for self-care	[39]
	Disease-related self-regulation	[33]
Self-efficacy	Disease-related self-efficacy	[28, 32, 39, 44, 46, 51, 55, 59, 63, 64, 86, 96, 109]
	Self-efficacy related to managing interactions with healthcare providers	[105, 86]
	General self-efficacy	[67, 86]
	Outcome expectations of disease-related self-management	[59]
Self-perception	Confidence regarding disease-specific management	[108]
	Self-rated competence in the areas of (a) scholastic work, (b) social acceptance, (c) athletic ability, (d) physical appearance, (e) behavioral conduct, and (f) global self-worth	[107]
Sense of control	Sense or locus of control	[63, 64]
	Role mastery	[110]

Appendix B. Overview of outcome measures used in the evaluation studies* (*continued*)

Groups of outcomes	Specific outcomes	References
Psychological outcomes	Depressive symptoms / depression	[51, 56, 61, 71, 74, 75, 78, 93-95]
	Anxiety or (dis)stress	[41, 62, 64, 71, 73, 74, 96]
	Catastrophic thinking about the condition	[74]
	Psychological symptoms	[89]
	Self-esteem	[39, 67, 71]
	Body image	[62]
Psychosocial functioning	Psychosocial adjustment	[29, 81]
	Developmental goals	[102]
	Psychosocial functioning	[49, 89, 99]
Coping	Coping or coping styles	[51, 63, 71]
	Adaptive and maladaptive behaviors and emotions	[42]
	Coping abilities/competencies and resourcefulness or coping styles	[40, 42, 44, 82]
	Internalizing and externalizing behavior	[61]
Health-related quality of life or subjective wellbeing	(Health-related) Quality of Life (generic)	[37, 54, 64, 69, 86, 93, 104]
	Disease-specific health-related quality of life	[27, 28, 30, 32, 33-36, 42-44, 46, 47, 51, 55, 56, 59, 61, 63, 76, 93, 96, 98]
	Subjective wellbeing	[110]
Vocational participation	School functioning (absence/attendance or grades)	[35, 43, 66, 73, 74, 105, 107, 109]
	Academic performance	[43]
	Occupational performance / work participation	[104]
	Pre-vocational experience	[98]
	Social and academic activities competencies	[61]
	Occupational problems	[102, 104]

Appendix B. Overview of outcome measures used in the evaluation studies* (*continued*)

Groups of outcomes	Specific outcomes	References
Social participation	Challenges / problematic situations encountered in different life areas	[70, 82]
	Days of changed plans	[35]
	Physical, emotional and behavioral concerns related to certain life areas	[62]
	(Autonomy in) participation in several life areas (e.g., living and romantic relationships)	[67, 72, 109]
	(Influence on) daily life with the condition	[47, 86]
	Social skills	[61, 92, 112]
	Social goals / goal attainment	[111, 112]
	Social acceptance by peers	[112]
	Dating	[62]
	Independence	[112]
	Parent involvement in diabetes care	[54]
	Family functioning	[51]
Family involvement or conflict (related to disease-related management tasks)	Division/sharing of responsibility, conflict or interaction within families related to disease specific tasks (e.g. blood glucose monitoring or insulin injections)	[46, 54, 57, 60, 85, 111, 112]
Support by others	Perceived support	[80]
	Social support by family and friends	[89]
Others (not included in groups)	Knowledge of the disease and/or treatment	[28, 32, 34, 36-39, 44, 49, 53, 62-64, 79, 82, 90, 92, 96, 98, 105, 107, 108, 111]
	Attitudes towards illness	[27, 78, 79, 86]
	Problem solving	[37, 78]

*Outcomes considering satisfaction with the intervention and proxy outcomes were not included in the analyses of outcome measures

3

Exploring effectiveness and effective components of self-management interventions for young people with chronic physical conditions: A systematic review

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ABSTRACT

Objective: To systematically explore the effectiveness and effective components of self-management interventions.

Methods: Study selection criteria were: Original articles in English published between 2003 and 2015; focusing on youth with chronic conditions; describing self-management interventions; with clear outcome measures; using RCT design. The random effects analysis was applied in which standardized mean differences per study were calculated.

Results: 42 RCTs were included. Interventions focused on medical management, provided individually in clinical settings or at home by mono-disciplinary teams showed a trend in improving adherence. Interventions delivered individually at home by mono-disciplinary teams showed a trend in improving dealing with a chronic condition.

Conclusions and practical implications: Adherence could be improved through interventions focused on medical management, provided individually in a clinical or home setting by a mono-disciplinary team. Interventions focused on dealing with a chronic condition might be provided individually, through telemedicine programs facilitating peer-support. These intervention elements seemed effective irrespective of diagnosis, and may therefore act as good starting points for further research into and for improvement of self-management support for youth with chronic conditions in pediatric care. Results underlined the need to systematically develop and evaluate self-management interventions, since this may provide more evidence for effectiveness and effective intervention components.

INTRODUCTION

Advancements in medical care result in increasing numbers of young people with chronic conditions making the transition into adulthood [1,2]. This transition, which implies becoming an independent adult fulfilling different roles in society, requires complicated life changes [3,4]. For these young people the transition may be more complicated, however, as they often lag behind in social and emotional development compared to healthy peers [5,6]. Furthermore, they are expected to take over the task of medical management from their parents, but this does not always come with ease [7,8]. It seems crucial, therefore, to help them develop self-management skills in all life areas [9-11]. Barlow et al. define self-management as “the individual’s ability to manage the symptoms and the consequences of living of a chronic, including treatment, physical, social, and lifestyle changes [12]”. As such, self-management and self-management support encompass three elements: medical management (considering the treatment), role management (considering participation in society), and emotion management (considering emotional consequences of being ill) [13]. This broad definition of self-management is widely used, as it fits with the World Health Organization’s definition of health in terms of physical, mental and social well-being [14].

A range of interventions is available for healthcare providers to support young people’s self-management in chronic disease [9-11,15,16]. These interventions are developed for young people aged between 7 and 25 years. The age of seven years is considered as a developmentally appropriate age to start the development of independence and young adults are still developing their full potential [17, 18, 19]. Self-management interventions have been reviewed in some studies [20,21], but solid evidence on effectiveness of self-management interventions (SMI) for young people with chronic conditions in general, i.e. irrespective of diagnosis, is still lacking [20-26]. Specifically, Kirk et al. [21] restricted themselves to asthma, cystic fibrosis and diabetes, while Lindsay et al. [20] restricted themselves to physical disabilities, excluding people with somatic chronic conditions. Moreover, although Kirk et al. [21] reviewed effective components of SMI for young people with asthma, cystic fibrosis and diabetes, insights into effective intervention components of self-management support for young people with a variety of chronic conditions are still lacking [20].

Still, looking at self-management support through a non-categorical approach is important [27]. Despite disease-specific differences, there are many commonalities between young people with chronic conditions, as they face comparable challenges and similar adaptive tasks while growing up [22]. A non-categorical approach to self-management support may benefit both health service delivery and research [28]. First, because

it could allow pediatric teams to learn from other specialist teams. Second, it could stimulate the use of same outcomes measures, which could enhanced the evaluation of effectiveness of these programs [28].

In a parallel paper including SMI evaluation studies using all kinds of study designs, a systematic overview of self-management support for youth with chronic conditions in general with respect to content, formats, theories, and evaluated outcomes is provided [29]. This systematic overview showed that most interventions were aimed at medical management, and that no theoretical base was provided for most of the included interventions. Although a variety of outcomes was evaluated, outcomes were conceptually related to specific content [29]. No results about effectiveness of SMI were included in this overview. Therefore, in the current paper, we reviewed published randomized controlled trials (RCT) of these SMI elaborating on a) evidence regarding effectiveness of SMI focused on youth with chronic conditions; and b) effective intervention components of these SMI, by employing a non-categorical approach to chronic conditions. Such insights may provide understanding of what elements of self-management support might be effective across a variety of chronic physical conditions, and hence may be valuable to further improve self-management support in health care.

METHODS

Study design

We applied a systematic review methodology which Grant and Booth defined as “systematically searching for, appraising and synthesizing research evidence, often adhering to guidelines for conducting a review” [30]. Methodological characteristics according to the ‘Search, Appraisal, Synthesis and Analysis’ (SALSA) framework are: exhaustive and comprehensive searching, quality assessment, narrative synthesis with tabular accompaniment, results of uncertainty around findings, recommendations for practice and future research [30]. This type of review allows for exploration of effects and effective intervention components of self-management support. The PRISMA statement guided the review process [31].

Search strategy

The search strategy employed variations and Boolean connections (AND, OR) of subject headings and keywords relating to self-management, children and adolescents, chronic illness, and intervention. For example, MESH terms (Medical Subject Headings) included: “Self care”, “Self medication”, “Intervention studies”, Program evaluation”. “Chronic disease”, “Disabled person”, “Adolescence”, “Child”. Key words included: “self help”; “clini-

cal effectiveness”; “evaluation”; “congenital disorder”; “handicapped child”; “youth”; “teenage”. Relevant variations of search terms were derived from database thesauruses and relevant review articles. An information specialist helped define the final search strategy. Six databases were searched: Embase; Medline; PsycINFO; Web-of-Science; CINAHL; and Cochrane. Two researchers (MB, JS) completed the database searches by scrutinizing relevant reviews’ references for additional relevant publications.

Inclusion criteria

- Study design: studies using a randomized controlled study design.
- Study types: original research articles in English language published from 2003 to April 2015, because the focus in literature on self-management rapidly increased since 2003 [32].
- Interventions: studies focusing on the evaluation of SMI and describing the SMI or referring to previous description(s) of the intervention.
- Outcome measures: studies considering clearly defined outcome measures.
- Participants: studies focusing on young people aged 7–25 years with somatic chronic conditions or physical disability. The age of seven years is considered a developmentally appropriate age to start the development of independence [17]. As young adults are still developing their full potential, the age range was extended from 18 to 25 years [18,19].

Studies had to meet all inclusion criteria to be included for further analysis.

Selection, quality assessment, and data extraction

Retrieved records (n = 7505) identified in databases and relevant reviews’ reference lists were imported into Endnote X7.2[®]. Two reviewers (MB, JS) independently categorized studies into: ‘include’, ‘exclude’ or ‘not clear’ based on title and abstract. Thereafter, the reviewers discussed the studies for which inclusion/exclusion was unclear and aimed to reach consensus on the decision. If doubt remained, a third reviewer was consulted (PR). Full texts of all agreed-upon articles (n = 562) were retrieved. Then, the two reviewers independently decided on the inclusion of articles based on the full text, resulting in 85 publications. For this paper we were interested in effects and effective intervention components, in contrast to the parallel paper [29], and therefore selected studies with RCT design (n=42) from the included publications. The selection process and reasons for exclusion are presented in Figure 1. Three reviewers (MB, JS, PR) independently assessed methodological quality of all included studies using the methodology checklists of the Scottish Intercollegiate Guidelines Network (SIGN) for randomized controlled trials [33]. Methodological quality was defined as high when at least eight out of ten criteria were met; as moderate when from five till eight were met; and as low when fewer than five

criteria were met [33]. Any discrepancies in assessment of methodological quality were resolved by discussion. Two reviewers (MB, JS) extracted data on study design; study sample; self-management domain considered during the intervention; interventions' theoretical base, format, discipline, interventionist, element, setting, outcome measures and study results [34]. Data was recorded on an electronic extraction form.

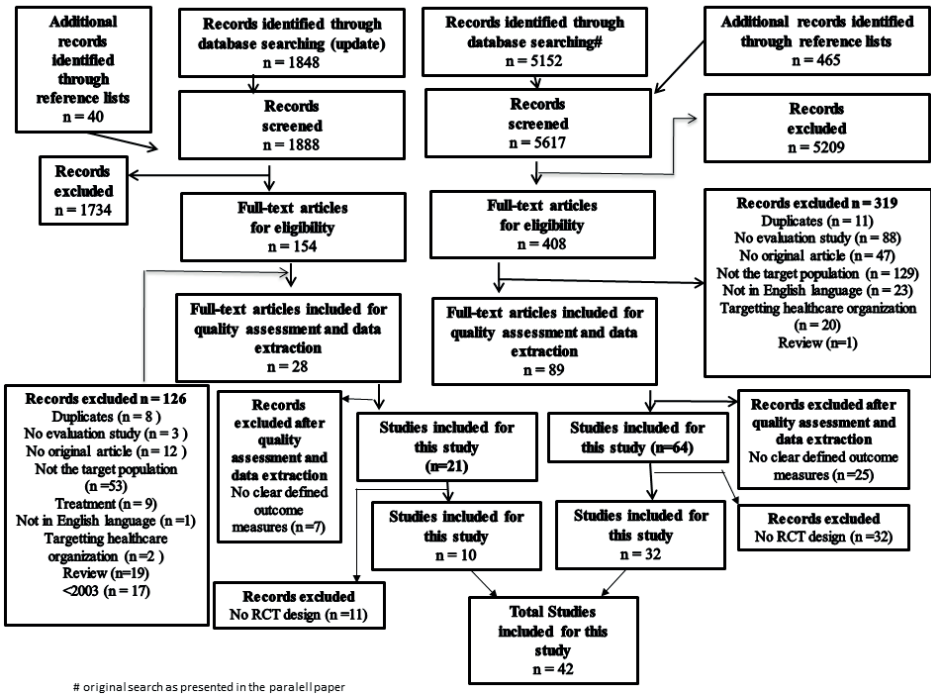


Figure 1. Selection process

Analysis

The following seven categories of outcome measures were inductively derived from the data: (1) symptoms; (2) disease knowledge; (3) adherence; (4) dealing with the chronic condition (including self-efficacy, coping and problem-solving); (5) psychological outcomes (including depression, stress, anxiety); (6) school attendance; and (7) quality of life. The reviewers (MB, JS) independently clustered the specific study outcome measures into these categories.

The overall quality of evidence for each outcome measure was independently rated by two researchers (MB&JS) using the GRADE approach, a method for rating the quality of evidence and grading the strength of recommendations per particular outcome [35]. Basically, evidence of randomized controlled trials (RCT's) is rated as high but may be

modified downwards on the basis of five different criteria: (1) risk of bias: assessment of methodological quality; (2) inconsistency: heterogeneity or variability in results across studies; (3) indirectness of evidence: indirect comparison of interventions within the studies; (4) imprecision of results: wide confidence intervals around the estimate of the effect; and (5) likeliness of publication bias: underestimating or overestimating of the effect due to the selective publication of studies [36-40]. Problems on a criterion was defined as serious when 60 - 80% of the studies showed limitations, and very serious when > 80% of the studies showed limitations on the criterion. The quality of evidence per outcome measure (high) was single downgraded for serious study limitations to moderate quality of evidence, and double downgraded for very serious study limitations to low quality of evidence.

Analyses to explore the effectiveness and the effective intervention components of self-management support were performed for each outcome measure separately. The random effects analysis was applied in which standardized mean differences per study were calculated [41]. In this way, we could compare effect sizes of statistically heterogeneous studies and provide an overview of effects on an outcome measure measured with different measurement instruments. Pooled estimates were not calculated, because interventions were clinically diverse, i.e. consisted of different intervention elements, and studies were statistically diverse, i.e. different instruments were used to evaluate the interventions. In addition, to evaluate the effective intervention components, we compared the effect sizes of included studies that differed on particular intervention components. For instance, effect sizes of individual intervention programs were compared with effect sizes of group intervention programs. Likewise, effect sizes of interventions focusing on different self-management domains, or provided by different disciplines, interventionists, and in various settings were compared. We speak of 'a trend in favor of', because we could not perform a meta-analysis (due to the heterogeneity in measurement instruments) and thus cannot state whether an intervention or its components are indeed effective or not. We only described a pattern of effects when *at least* three studies were included, and used the cut-off point of two out of three studies (67%) as a minimum number of studies indicating effects in the same direction. Otherwise, the pattern was described as showing no clear effects. When drawing conclusions, we downgraded the level of evidence for those issues where the number of available studies was low [38]. Individual studies that showed a significant effect were weighted twice and individual studies that showed a trend (but not a significant effect) were weighted once. Review Manager version 5.1 software was used for the analyses.

RESULTS

Paralell paper

A detailed overview of the focus and content of the interventions presented in the parallel paper, showed a variety in the operationalization of self-management support [29]. Most SMI were aimed at medical management, neglecting psychosocial challenges. Although a variety of outcomes was evaluated, outcomes were conceptually related to specific content [29]. In order to explore the effectiveness and effective components of SMI, we selected the studies with RCT-design from the included studies (n=42). In the current paper results on effectiveness and effective components of SMI are described.

General study characteristics (n = 42)

- Countries: Most, studies were performed in the USA (n=27), followed by Australia (n=3), Canada (n=2), Taiwan (n=2), the Netherlands (n=2), the United Kingdom (n=2), China (n=1), Germany (n=1), and Jordan and Australia (n=1).
- Chronic conditions: Study participants were patients with asthma (n=16), diabetes (n=6), cystic fibroses (n=3), cancer (n=2), human immunodeficiency virus infection (n=2), sickle cell disease (n=2), or other diseases (e.g., spina bifida, haemophilia and juvenile fibromyalgia) (n=9).
- Methodological quality: The methodological quality of 25 studies was rated as moderate, of 11 studies as high and of 6 studies as low.

Intervention components (n=42)

Many interventions were solely aimed at medical management (61.9%). Medical management was either disease-specific or more general. Disease-specific medical management refers to tasks or topics associated with or related to a specific diagnosis, e.g., self-monitoring of blood glucose values in diabetes. General medical management refers to health and healthcare related tasks irrespective of diagnosis. For instance, accessing healthcare, but also child-parent sharing or teamwork related to medical management tasks. One intervention considered only role management (2.4%), referring to topics related to social participation, such as communicating, assertiveness and keeping up with peers. None of the interventions considered emotion management separately. Interventions considering emotion management could refer to young person's feelings and intrinsic characteristics. Topics covered are stress-management and relaxation techniques. The other interventions addressed multiple domains, for example both on medical and role management (35.7%). Studies did not mention a theoretical base for the intervention (64.3%), referred to learning theories like Bandura's (cognitive) social learning theory (11,9%), cognitive behavioral theory (4,4%) or a combination of these (2,4%). Interventions were either applied at individual level (69.0%), at group level

(23.8%) or both (7.1%). Most interventions were provided by a mono-disciplinary team (70%). Interventions included education (88.1%), peer-support (23.8%), problem solving (23.8%), relaxation training (21.4%), cognitive restructuring (e.g. cognitive behavioral therapy) (19.0%), or self-monitoring, (e.g. keeping a diary with blood glucose values) (19.0%). In general, neither the domain of self-management considered during interventions nor the other intervention elements were specifically linked to certain theoretical frameworks. Intervention settings were inpatient or outpatient clinics (31.0%), home (23.8%), online (14.3%), school (14.3%), home and clinic (7.1%), camping sites (4.8%), or home and school (2.3%). Settings were not exclusive for formats and elements of interventions.

A detailed description of study characteristics and intervention components per study is presented in Appendix A.

Exploration of effectiveness and effective components of self-management interventions

Outcome measure symptoms

Twenty-four studies focused on the severity of symptoms as an outcome measure [43, 49, 51, 54, 56, 58-60, 63-67, 70, 72, 74, 76-83]. In general, SMI showed a trend in favor of the control condition. Specifically, nine studies showed a trend in favor of intervention; twelve studies reported a trend in favor of the control condition; and three studies showed a significant effect in favor of the control condition. The quality of evidence was very low (Table 1).

Of all interventions that aims to reduce the severity of symptoms, the interventions focused on medical management, medical and role management, or medical management, emotion management and role management showed a trend in favor of the control condition. The same pattern held for interventions delivered by a psychologist. Also, interventions provided in a clinic, online, or at home and in a clinic showed a trend in favor of the control condition. In contrast, interventions provided at home showed a trend in favor of intervention (Figures 2a-2c).

Table 1. Evidence profile

Quality assessment					Number of patients	Quality of the evidence
Outcome (No of studies)	Limitation in design	Inconsistency	Indirectness	Imprecision		
Symptoms (24)	No serious limitation	Serious inconsistency	No serious indirectness	Very serious imprecision	Unlikely	1019 1085 Very low
Disease knowledge (19)	No serious limitation	Serious inconsistency	No serious indirectness	No serious imprecision	Unlikely	771 626 Moderate
Adherence (9)	No serious limitation	Serious inconsistency	No serious indirectness	Very serious imprecision	Unlikely	333 387 Very low
Dealing with chronic condition in daily life (11)	No serious limitation	Very serious inconsistency	No serious indirectness	Serious imprecision	Unlikely	395 535 Very low
Psycho-logical outcomes (9)	No serious limitation	Serious inconsistency	No serious indirectness	Very serious imprecision	Unlikely	367 378 Very low
School atten-dance (5)	Serious limitation	Serious inconsistency	Very serious indirectness	No serious imprecision	Unlikely	305 285 Very low
Quality of life (13)	No serious limitation	No serious inconsistency	No serious indirectness	No serious imprecision	Unlikely	705 757 High

^a number of patients that participated in the control condition.
^b number of patients that participated in the intervention condition

Exploring effectiveness and effective components of self-management interventions

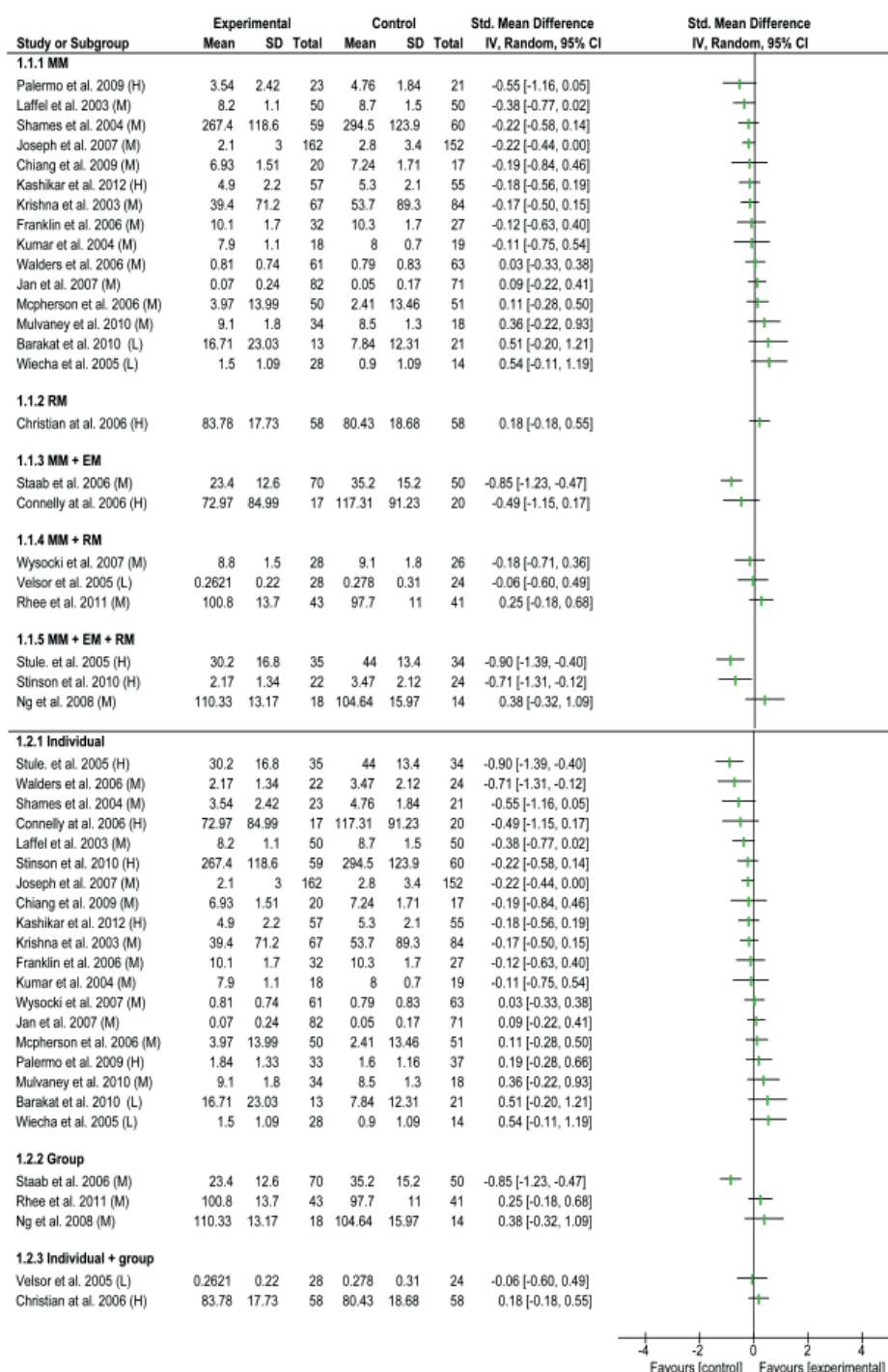


Figure 2A. Effectiveness of intervention component domain of self-management and format on symptom reduction

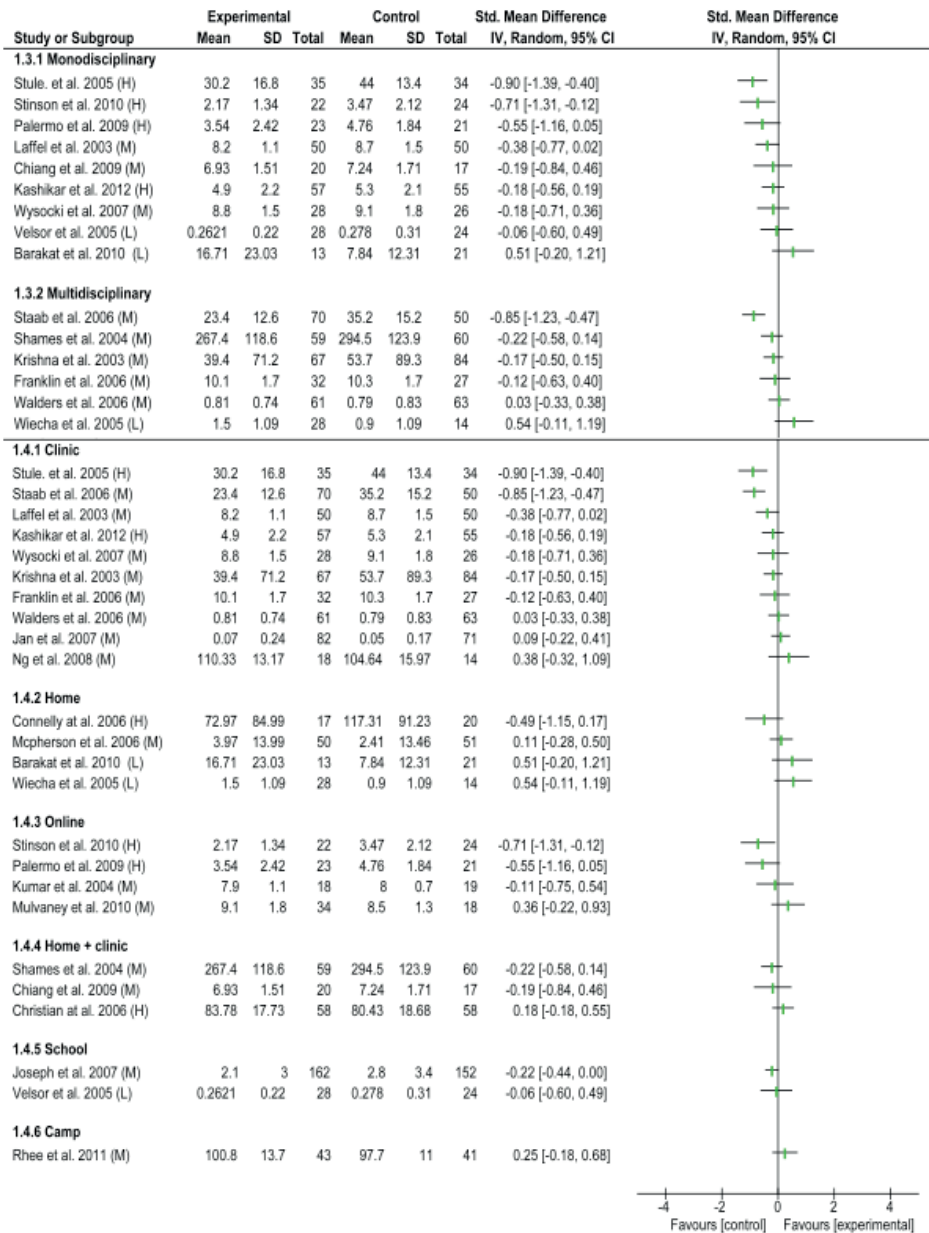


Figure 2B. Effectiveness of intervention component discipline and setting on symptom reduction.

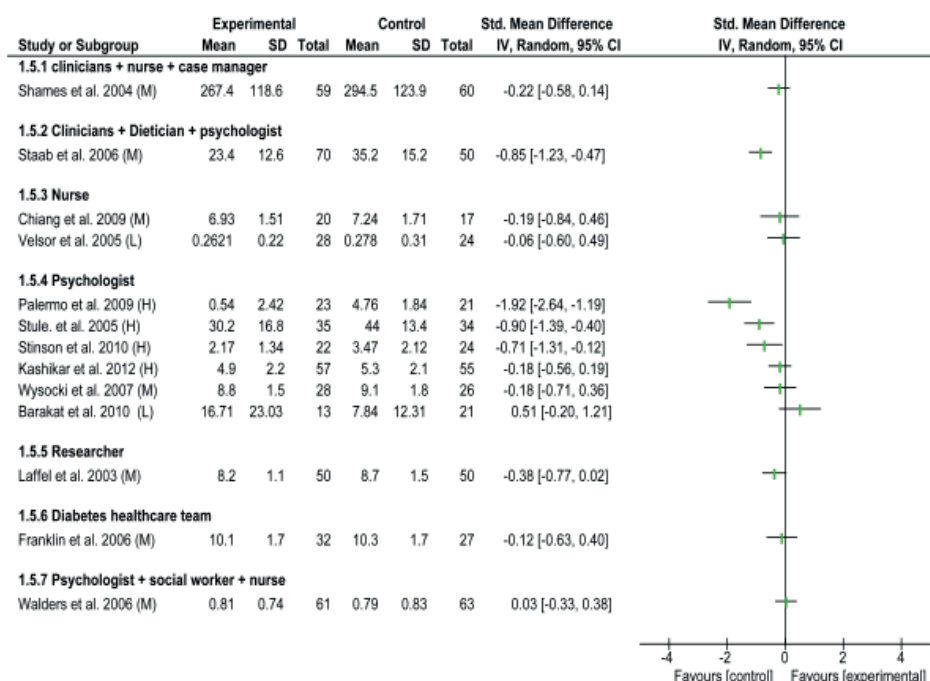


Figure 2C. Effectiveness of intervention component interventionists on symptom reduction.

Outcome measure disease knowledge

Nineteen studies focused on disease knowledge as an outcome measure [42, 43, 45, 47, 48, 52-55, 57, 61-63, 66, 71, 73, 76, 78, 82]. In general, SMI showed a trend in favor of intervention. Specifically, nine studies revealed a significant effect in favor of intervention; five studies showed a trend in favor of intervention and five studies showed a trend in favor of the control condition. The quality of evidence was moderate (Table 1).

Of all interventions that aims to improve disease knowledge, interventions focused on medical management, or medical management, emotional management and role management, showed a trend in favor of intervention. The same pattern revealed for interventions provided individually and interventions provided in a clinic or at home. Mono-disciplinary interventions showed a trend in favor of the control condition, but this was also the case for multi-disciplinary interventions (Figures 3a-3b).

Outcome measure adherence

Nine studies focused on adherence as an outcome measure [45, 53, 54, 61, 67, 68, 78, 82, 83]. Overall, SMI showed a positive trend on adherence. Specifically, four studies showed a significant effect in favor of intervention, five other studies showed a positive tendency. The quality of evidence was very low (Table 1).

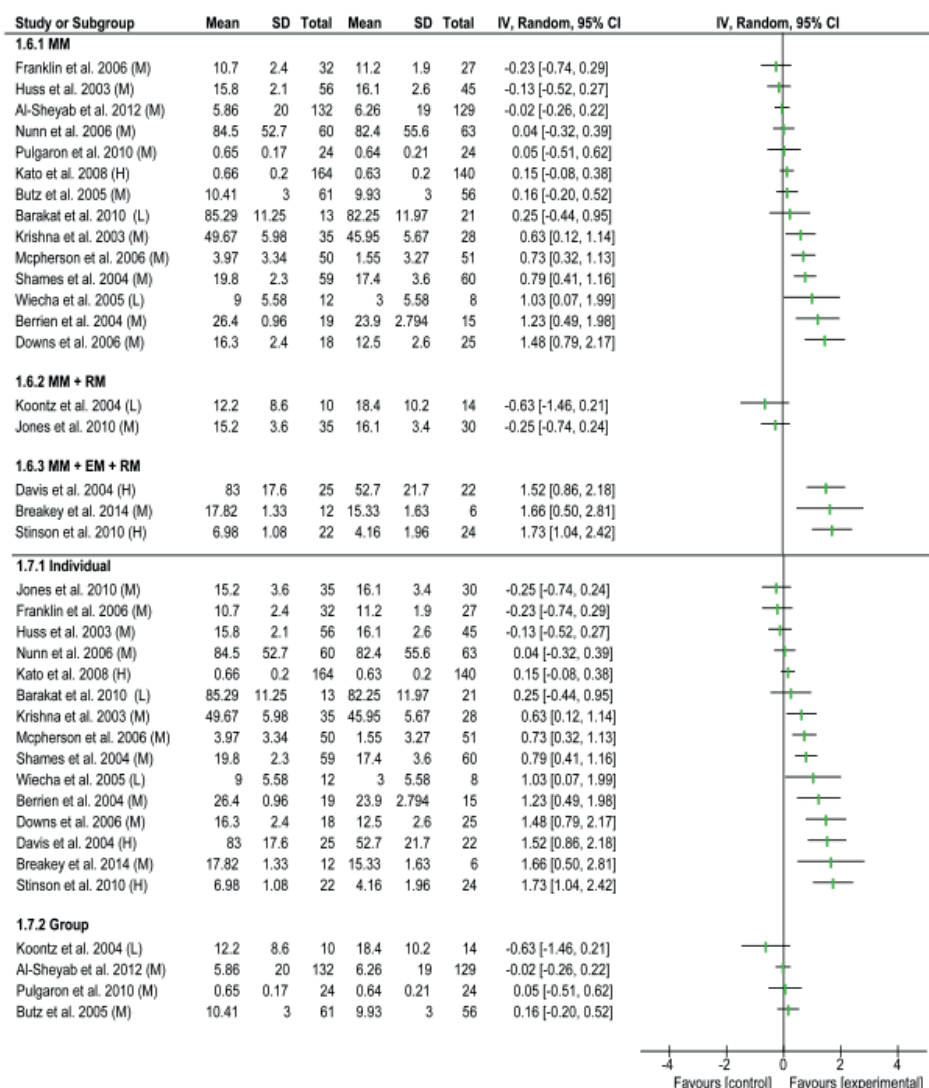


Figure 3A. Effectiveness of intervention component domain of self-management and format on disease knowledge.

Exploring effectiveness and effective components of self-management interventions

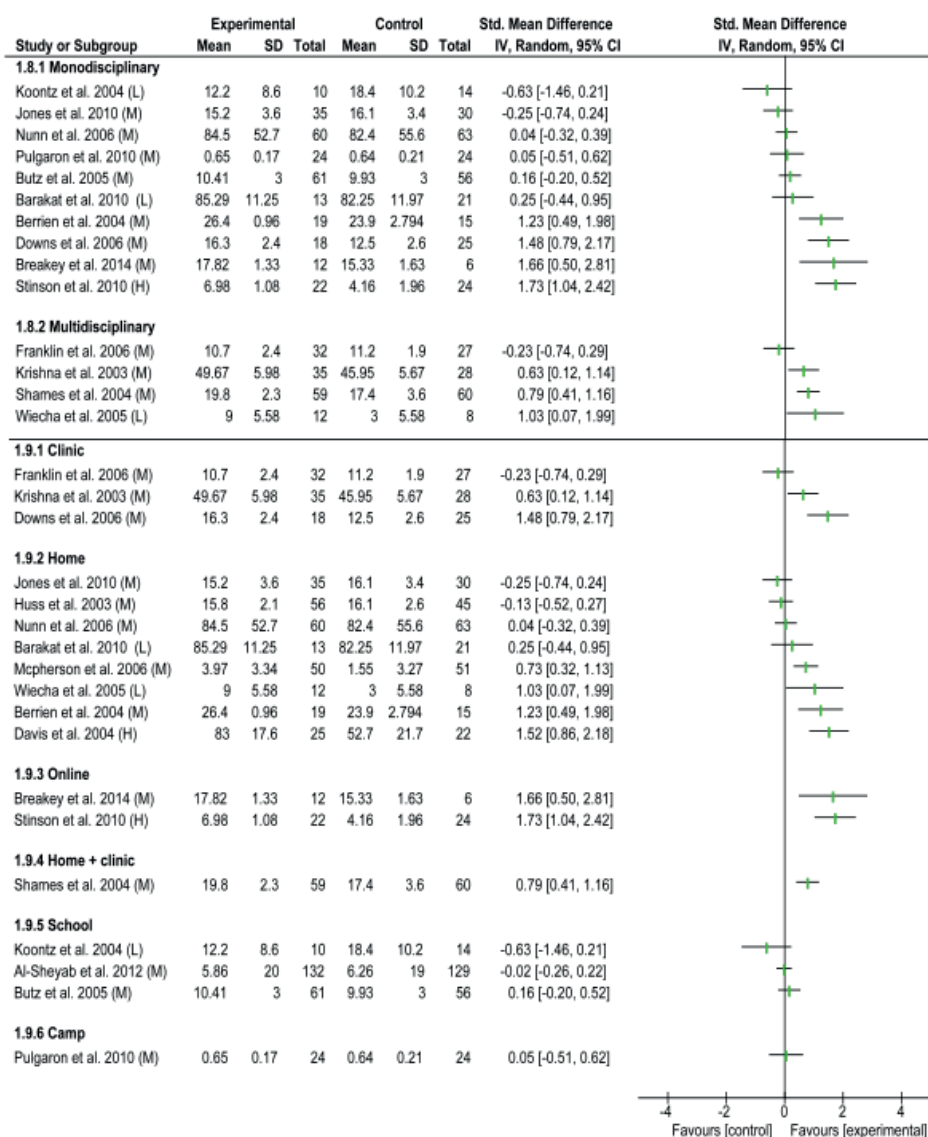


Figure 3B. Effectiveness of intervention component discipline and setting on disease knowledge.

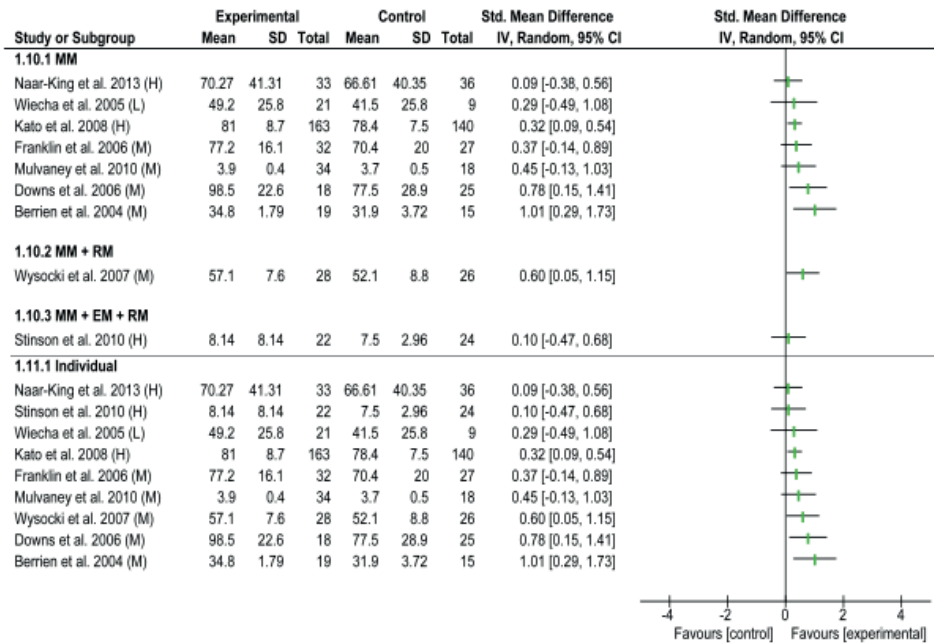


Figure 4A. Effectiveness of intervention component domain of self-management and format on adherence.

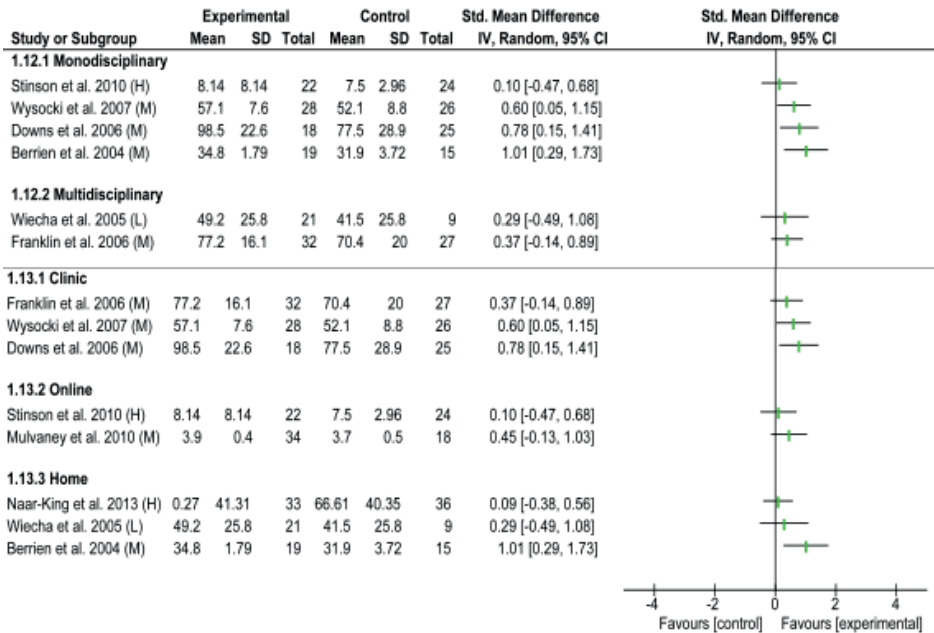


Figure 4B. Effectiveness of intervention component discipline and setting on adherence.

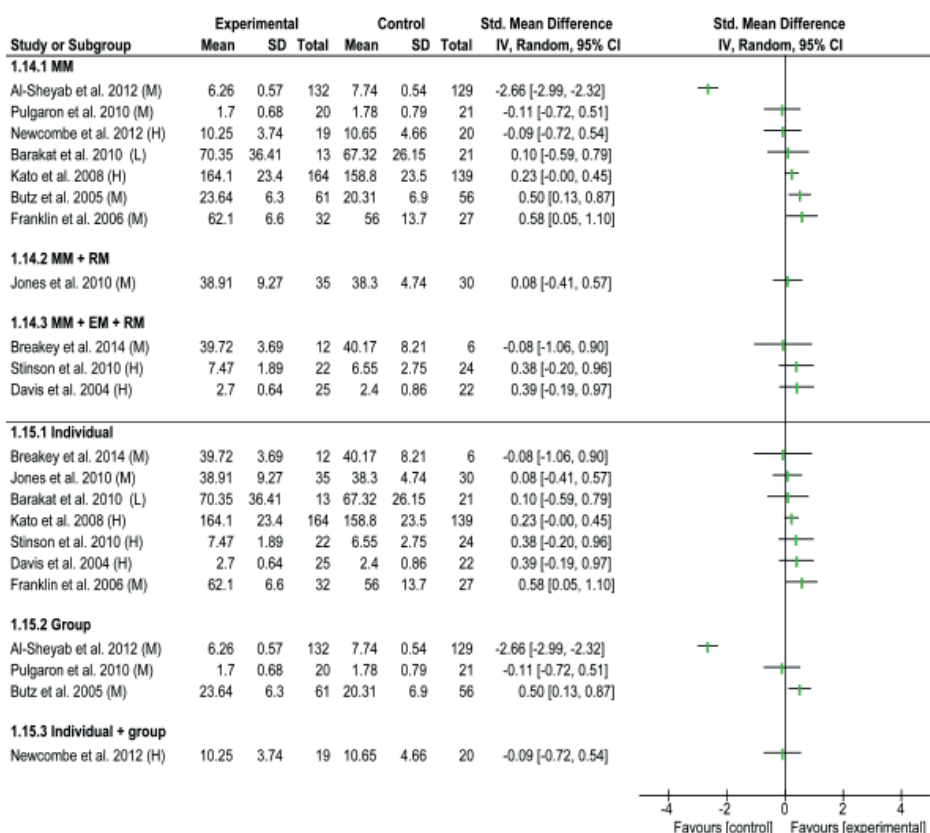


Figure 5A. Effectiveness of intervention component domain of self-management and format on dealing with a chronic condition in daily life.

Of all interventions that aims to improve adherence, interventions focused on medical management showed a trend in favor of intervention. The same pattern held for interventions provided individually and in a clinic or at home and interventions delivered mono-disciplinary (Figures 4a-4b).

Outcome measure dealing with the chronic condition in daily life

Eleven studies focused on dealing with the chronic condition in daily life as an outcome measure [42, 43, 47, 48, 52, 54, 57, 61, 69, 73, 78]. Overall, no clear effects of SMI were found on dealing with the chronic condition in daily life. Specifically, two studies had a significant effect in favor of intervention; five studies had a trend in favor of intervention. In contrast, three studies revealed a trend in favor of the control condition and one study showed a significant effect in favor of the control condition. The quality of evidence was very low (Table 1).

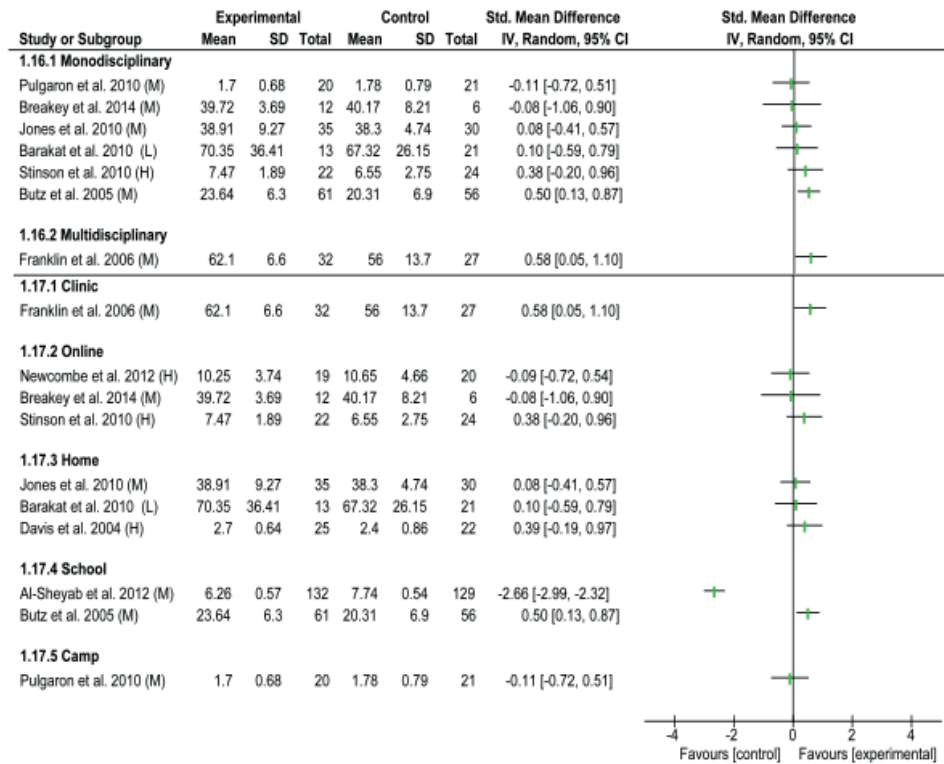


Figure 5B. Effectiveness of intervention component discipline and setting on dealing with a chronic condition in daily life.

Of all interventions that aims to improve dealing with chronic condition in daily life, interventions focused on medical management, showed a trend in favor of intervention. The same pattern held for mono-disciplinary interventions provided individually and at home. In contrast, interventions provided online showed a trend in favor of the control condition (Figures 5a-5b).

Outcome measure psychological outcomes

Nine studies focused on psychological outcomes [44, 46, 49, 59-61, 69, 72, 75, 78]. In general, no clear effects of SMI were found on this outcome measure. Specifically, one study had a significant effect in favor of intervention; two studies showed a positive tendency; and six others showed a trend in favor of the control condition. The quality of evidence was very low (Table 1).

Of all interventions that aims to improve psychological outcomes, interventions focused on medical management, showed a trend in favor of the control condition. The same pattern was revealed for mono-disciplinary interventions and interventions provided

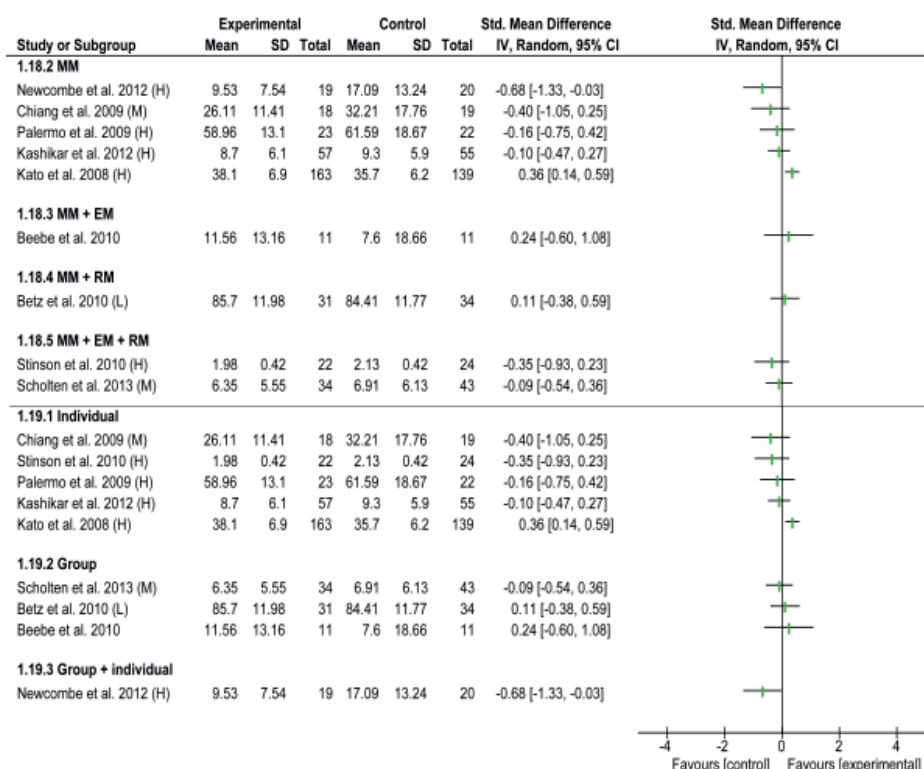


Figure 6A. Effectiveness of intervention component domain of self-management and format on psychological outcomes.

individually by a psychologist and online. In contrast, interventions provided in groups showed a trend in favor of intervention (Figures 6a-6c).

Outcome measure school attendance

Five studies focused on school attendance as an outcome measure [43, 58, 62, 63, 79]. SMI showed a trend in favor of the control condition: two studies showed a significant effect in favor of the control condition one study showed a negative tendency, while two studies showed a trend in favor of intervention. The quality of evidence was very low (Table 1).

No clear pattern was revealed for intervention components.

Outcome measure quality of life

Thirteen studies focused on quality of life as an outcome measure [42, 44, 47-50, 57, 58, 61, 65, 70, 74, 78]. Overall, no clear effects of SMI were found on quality of life. Four studies had a significant effect in favor of intervention; three studies showed a trend

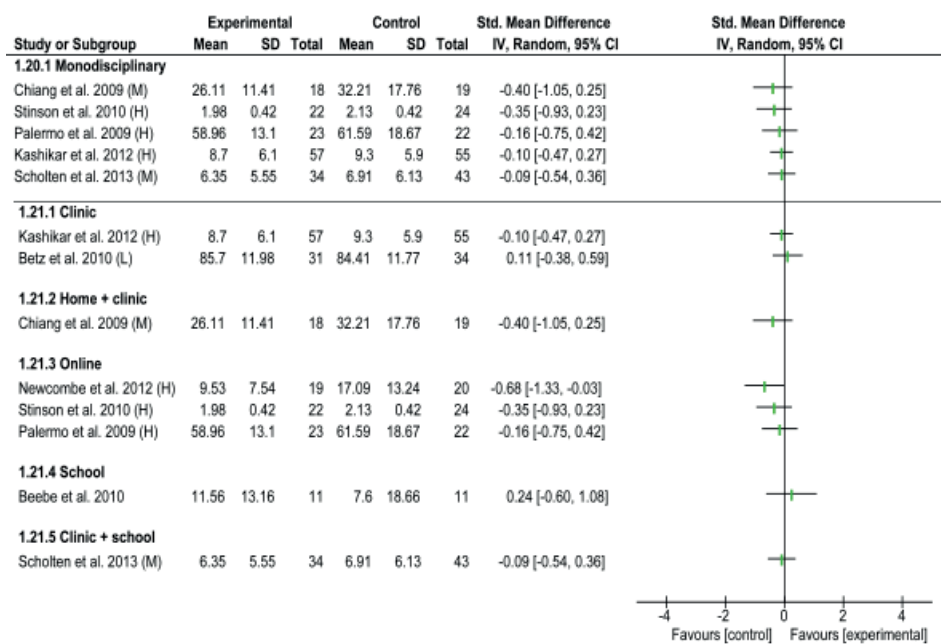


Figure. 6B Effectiveness of intervention component discipline and setting on psychological outcomes.

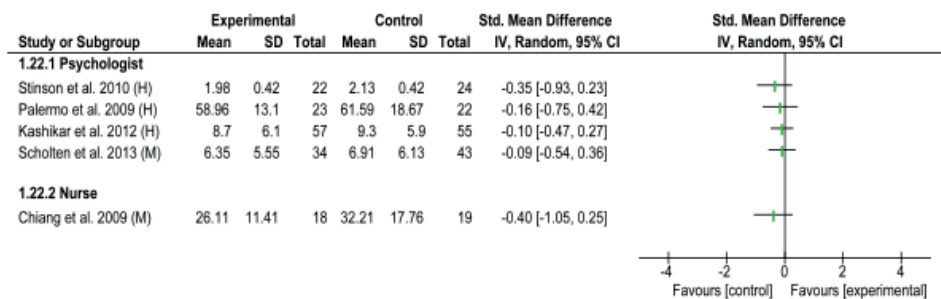


Figure 6C. Effectiveness of intervention component interventionists on psychological outcomes.

in favor of intervention; one study showed a significant effect in favor of the control condition; four studies showed a trend in favor of the control condition. The quality of evidence was high (Table 1).

Of all interventions that aims to improve quality of life, interventions focused on medical management showed a trend in favor of intervention (Figure 7).

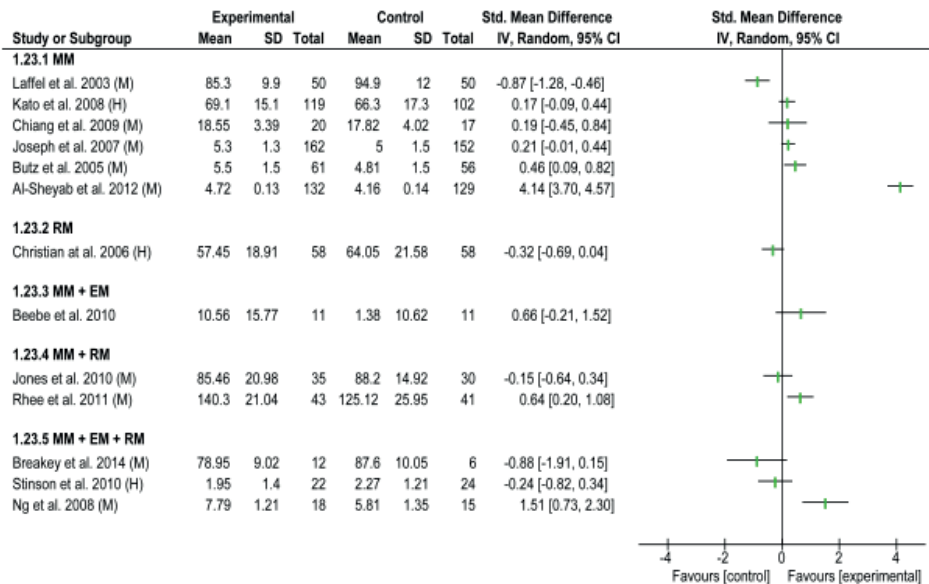


Figure 7. Effectiveness of intervention component domain of self-management on quality of life

DISCUSSION

We reviewed randomized controlled trials to explore the effectiveness and effective intervention components of SMI. In contrast to existing literature, we looked at pediatric SMI in general, i.e. irrespective of type of condition. Our findings suggests that pediatric SMI might be effective at influencing disease knowledge, adherence, but not at symptom severity and school attendance. Conflicting evidence was found for effectiveness of SMI on dealing with the chronic condition, psychological outcomes and quality of life. However, results about effectiveness should be interpreted with some caution, because the quality of evidence for most outcome measures was rated as low. In addition, some evidence was found for effective intervention components, but this differs per outcome measure.

Effectiveness of SMI

Possible evidence for effectiveness of SMI on disease knowledge was also reported in previous reviews on SMI for adult patients [12,84]. Others showed possible evidence for effectiveness of SMI on disease knowledge of youth with spina bifida, arthritis, asthma, or diabetes [17,18,85-87] and for effectiveness of pediatric SMI on adherence [88-90]. Moreover, earlier studies on youth showed that both knowledge about the condition and medication adherence are important to maintain health [91].

Although Kirk et al. [21] underlined the need to incorporate patient-centered outcomes such as quality of life and psychosocial well-being into SMI evaluation studies, conflicting evidence for effectiveness of SMI on these outcome measures was found and inconsistent results have been reported by others [12,84]. Nolte et al. [92] pointed out that patient reported outcomes measures (e.g. quality of life and psychosocial well-being) are highly variable, since these require the highest degree of personal appraisal. Specifically, people may define depression or quality of life very differently before SMI compared to after intervention, influencing the reliability of measurements. These biases could have influenced the results of included studies and, thus, our results. However, patient reported outcomes do provide insights into patients' lived experiences and are therefore highly relevant for studying self-management. Therefore, future studies designed to explore response shifts are recommended. For instance, qualitative methods may shed light on how a patient's context interacts with their lived experiences. This is in line with the recently posed notion that patient's context is important to consider when researching self-management [93].

Effective components of SMI

In addition to previous research on effectiveness of SMI [17,18,21], this review revealed some suggestions about components of SMI to be effective. Specifically, SMI aimed to improve adherence might be effective when focused on medical management and provided individually in a clinic or at home by a mono-disciplinary team. This is not surprising, since adherence is part of medical management and individual attention of one professional may enhance insights into personal barriers to adherence.

We found some evidence that SMI aiming to support young people to deal with the chronic condition in daily life should focus on medical management, emotion management and role management, i.e. should consider a holistic view on self-management [13, 22]. This conclusion is in line with earlier findings showing that one's illness behavior depends on one's perception of symptoms and on the extent to which symptoms interfere with daily life [94], i.e. one's lived experiences [95, 96]. In this light, the support need on dealing with chronic conditions in daily life is influenced by lived experiences, and SMI on this outcome measure should therefore include role management and emotion management, besides medical management [13]. In addition, our review suggests that these SMI might be mono-disciplinary programs, provided individually and at home. These findings are somewhat unexpected since peer-support was found to be an appropriate element to stimulate dealing with the chronic condition in daily life, i.e. enhance self-efficacy, and therefore we expected group sessions and a clinical setting to be effective intervention elements [97,98]. However, taking a closer look at elements used in these SMI, we conclude that these individually provided programs facilitate peer-support us-

ing telemedicine element. This indicates that online peer-support could also be an appropriate element to improve self-efficacy, problem solving skills and pro-active coping behavior, as was earlier mentioned in literature [18]. However, more research is needed to further validate this hypothesis, since we found some evidence that interventions provided online without a peer-support element might not be effective.

Our review suggests, too, that a mono-disciplinary team, an online setting and an individual format might not be effective intervention components of self-management support focused on psychological outcomes, i.e. stress, anxiety and depression. In contrast, we found some evidence for group sessions to be an effective intervention component. Earlier research showed the perceived distress of people with chronic conditions and their need for social support [99]. It might be necessary, therefore, to incorporate elements that promote people's sense of empowerment into SMI, if aiming to improve psychological outcomes [100,101]. In addition cognitive behavioral therapy is commonly used in the treatment of depression. According to the rationale behind the therapy, cognitive social learning theory, people should adapt their maladaptive cognitions and modify their behavior to reduce the depressive symptoms [102]. Group meetings may help to discuss maladaptive cognitions and behaviors, and may help to change these into more adaptive ones. Also, our review suggests that SMI aimed to target psychological outcomes might not be focused on medical management. This is not unexpected, because emotion management or a combination of emotion management with role and/or medical management might be a more appropriate content for SMI aiming to reduce stress, anxiety and depression.

Furthermore, we found that programs on symptom reduction should be provided at home instead of in a clinic, home and clinic or online. An explanation for this finding could be that symptoms arise in interacting with the social context. In line with this reasoning, SMI could be more effective when provided in people's daily life. For example, Reeves et al. [103] indicated the importance of social community to fulfill self-management tasks. However, Krieger et al. [104] reviewed pediatric asthma interventions and found both home and clinical settings to be effective for symptom reduction. Also, no clear effects were found for any other intervention components. Elements of SMI on symptom reduction seem to be too diverse and sample sizes are too small, which hampers the evaluation of working elements of these interventions. It seems that the development of SMI is often not based on theories [21, 24, 25, 29]. Therefore, it is recommended to further standardize the development of SMI, and thus to explicitly use theoretical frameworks when designing interventions. Also, it could be that symptoms are too disease-specific and therefore effects on symptom reduction could not be appropriately analyzed in a review of SMI focusing on a variety of chronic conditions. Therefore it would be favor-

able to employ a disease specific approach, when exploring effects of SMI on symptom reduction.

Strengths limitations and other considerations

SMI for young people across the wide age range of 7-25 years were included, while content or themes obviously are not applicable to the whole age range, e.g. vocational participation pertains to older adolescents only. It would be advisable, therefore, to distinguish between age groups. However, a systematic overview of the content of SMI for young people aged between 7 and 25 years showed that interventions' theoretical base, formats and elements did not differ for different age groups [29].

This study looked at many types of SMI across a wide range of chronic conditions. The breadth of this study is a potential strength. The findings enable researchers and health-care professionals to look at general self-management support, which goes beyond particular chronic conditions. In addition, subgroup analyses -not presented in this paper- showed no differences in effects between interventions focused on a distinctive diagnosis.

Last, the, in general, low quality of evidence for most outcome measures as rated using the GRADE methodology [35], downgrades the strength of recommendations per outcome measure. The low quality of evidence was mainly caused by the variability and heterogeneity of study results. Also it was not possible to perform a meta-analysis on the effects of SMI, since the outcome measures and intervention programs were very heterogeneous. Therefore, results about effectiveness should be interpreted with some caution. Different conceptualizations of self-management might be one explanation for the diversity of elements and content of SMI and the variety of outcome measures used in SMI evaluation studies, which points at a more fundamental challenge to provide precise definitions [105]. Others, too, recommended to further standardize the development and evaluation of self-management interventions [16,17,84]. Intervention mapping methodology [98] is a recommended stepwise approach for theory and evidence-based development and implementation of interventions that helps to fit intervention goals with program development and program evaluation. In addition, a theoretically derived intervention would make for a more effective SMI, using components that have been found to be effective in changing that specific outcome [106].

CONCLUSION

Our review suggests that pediatric SMI might be effective at influencing disease knowledge and adherence across a wide range of conditions. There are indications that SMI aimed to improve *adherence* should be focused on *medical management*, and should be provided *individually in a clinical setting or at home by a mono-disciplinary team*. Furthermore, a *holistic view on self-management, individual format and home setting* combined with *online peer-support* might be effective intervention components for SMI focused on *dealing with the chronic condition* in daily life. These combinations of expected outcomes, focus and intervention elements seemed effective irrespective of diagnosis, and may therefore act as good starting points for further improvement of and research into self-management support of young people with chronic conditions in health care. Results underlined the need to systematically develop and evaluate self-management interventions, since this may provide more evidence for effectiveness and effective intervention components.

PRACTICAL IMPLICATIONS

Self-management support is important for people with chronic conditions to help them deal with their condition in daily life. This is even more pertinent to young people growing up with chronic conditions, who have to face the normal tasks of development (e.g. acquiring autonomy) and have to engage in lifelong medical management of their condition. Our systematic overview of effectiveness and effective intervention components may assist clinicians and other healthcare professionals in determining the focus, content and components of self-management support they provide. More specifically, to improve patient's adherence, healthcare professionals should focus their SMI on medical management, and they should provide the intervention individually in a clinical setting or at home by a mono-disciplinary team. Furthermore, a holistic view on self-management, individual format and home setting combined with online peer-support might be appropriate intervention components to select for SMI focused on dealing with the chronic condition in daily life.

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Declaration of interest

The authors have no potential conflicts of interests to declare, including relevant financial interests, activities, relationships, and affiliations.

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Appendix A. Study characteristics and intervention components

Background		Participants		Study characteristics		Components of self-management interventions						
Studies:	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base ^a (Yes) ^b	Domain of self-management ^b	Formats	Elements	Interventionists	Setting
Al-Sheyab et al. 2012 [42]	Jordan	Asthma	N = 261 I = 132 C = 129	Moderate	Usual care	Disease knowledge	NA	MM	Group	Education	Peers	School
	Australia					Quality of life				Discussion		
						Dealing with the chronic condition in daily life				Problem-solving skills		
Barakat et al. 2010 [43]	USA	Sickle cell disease	N = 37 I = 17 C = 20	Low	Another specific intervention	Symptoms	NA	MM	Individual	Education	Psychologist	Home
						Dealing with the chronic condition in daily life				Cognitive restructuring		
						School attendance				Self-monitoring		
Beebe et al. 2010 [44]	USA	Asthma	N = 22	Low	Waitlist	Disease knowledge				Relaxation training		
						Quality of life				Art-therapy		
Berrien et al. 2004 [45]	USA	HIV	N = 3 I = 20 C = 17	Moderate	Usual care	Psychological outcomes	NA	MM+EM	Group	Education	NA	School
						Disease knowledge	NA	MM	Individual	Education	Nurse	Home
						Adherence						

Background		Participants		Study characteristics			Components of self-management interventions						
Studies:	Country	Diagnosis	N	Methodological Quality		Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventionists	Setting
				Low	Moderate								
Betz et al. 2010 [46]	USA	Spina Bifida	N = 65 I = 31 C = 34	Low	Moderate	Usual care	Psychological outcomes	NA	MM + RM	Group	Goal-setting skills	NA	Clinic
Breakey et al. 2014 [47]	Canada	Haemo-philia	N = 29 I = 16 C = 13	Moderate	Usual care	Usual care	Quality of life	NA	MM + EM + RM	Individual	Education	Research assistant	Online
							Dealing with the chronic condition in daily life				Relaxation training		
							Disease knowledge				Peer-support		
Butz et al. 2005 [48]	USA	Asthma	N = 200 I = 112 C = 98	Moderate	Usual care	Usual care	Quality of life	NA	MM	Group	Education	Asthma educator	School
							Dealing with the chronic condition in daily life				Telemedicine		
							Disease knowledge						

Background		Participants		Study characteristics		Components of self-management interventions							
Studies:	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventionists	Setting	
Chiang et al. 2009 [49]	Taiwan	Asthma	N = 48 I = 22 C = 26	Moderate	Another specific intervention	Quality of life	NA	MM	Individual	Education	Nurse (undergraduate degree)	Home	
Christian et al. 2006 [50]	USA	Cystic Fibrosis	N = 116 I = 58 C = 58	High	Usual care	Quality of life	NA	RM	Individual	Education	NA	Home Clinic	
						Symptoms				Group			Problem solving skills
													Peer-support
Connelly et al. 2006 [51]	USA	Mi-graine	N = 31 I = 14 C = 17	High	Waitlist	Symptoms	NA	MM+EM	Individual	Education	NA	Home	
Davis et al. 2004 [52]	USA	Cystic Fibrosis	N = 47 I = 25 C = 22	High	Waitlist	Dealing with the chronic condition in daily life	NA	MM+ RM+ EM	Individual	Relaxation training	NA	Home	
													Cognitive restructuring
													Education
						Disease knowledge							

Background		Participants		Study characteristics		Components of self-management interventions						
Studies:	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventionists	Setting
Downs et al. 2006 [53]	Australia	Cystic Fibrosis	N = 43 I = 18 C = 25	Moderate	Not clear	Adherence	CSL	MM	Individual	Education	Healthcare professional	Clinic
						Disease knowledge				Problem-solving skills		
										Decision-making skills		
Franklin et al. 2006 [54]	UK	Diabetes	N = 90 I1 = 32 I2 = 31 C = 27	Moderate	Another specific intervention	Symptoms	CSL	MM	Individual	Telemedicine	Diabetes healthcare team	Clinic
						Dealing with the chronic condition in daily life				Goal-setting skills		
						Adherence						
Huss et al. 2003 [55]	USA	Asthma	N = 101 I = 56 C = 45	Moderate	Usual care	Disease knowledge	NA	MM	Individual	Education	NA	Home
Jan et al. 2007 [56]	Taiwan	Asthma	N = 164 I = 88 C = 76	Moderate	Another specific intervention	Symptoms	NA	MM	Individual	Education	NA	Clinic (out-patient)
										Self-monitoring		

Background	Participants	Study characteristics			Components of self-management interventions							
Studies:	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventionists	Setting
Jones et al. 2010 [57]	USA	Cancer	N = 65 I = 35 C = 30	Moderate	Another specific intervention	Quality of life Dealing with the chronic condition in daily life	NA	MM+RM	Individual	Education	Healthcare professional	Home
Joseph et al. 2007 [58]	USA	Asthma	N = 314 I = 162 C = 152	Moderate	Another specific intervention	Disease knowledge Quality of life Symptoms School attendance	Trans-theoretical/Health belief model	MM	Individual	Education	NA	School
Kashikar-Zuck et al. 2005 [59] Kashikar-Zuck et al. 2012 [60]	USA	Juvenile Fibro-myalgia	N = 114 I = 57 C = 57	High	Another specific intervention	Symptoms Psychological outcomes	CBT	MM	Individual	Cognitive restructuring Education Relaxation training Problem-solving skills	Psychologist	Clinic

Background		Participants		Study characteristics		Components of self-management interventions						
Studies:	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventionists	Setting
Kato et al. 2008 [61]	USA	Cancer	N =371 I = 195 C = 176	High	Another specific intervention	Quality of life	CSL	MM	Individual	Education	NA	NA
						Dealing with the chronic condition in daily life						
						Psychological outcomes						
						Adherence						
Koontz et al. 2004 [62]	USA	Sickle cell disease	N = 24 I = 10 C = 14	Low	Another specific intervention	Disease knowledge						
						School attendance	NA	MM+RM	Group	Education	Teachers	School
						Disease knowledge				Peer-support		
Krishna et al. 2003 [63]	USA	Asthma	N = 228 I = 107 C = 121	Moderate	Another specific intervention	Symptoms	NA	MM	Individual	Education	Multi-disciplinary team	Clinic
						School attendance				Decision-making skills		
						Disease knowledge				Social skills		
Kumar et al. 2004 [64]	USA	Diabetes	N = 40 I = 19 C = 21	Moderate	Another specific intervention	Symptoms	NA	MM	Individual	Self-monitoring	NA	Online

Background	Participants		Study characteristics		Components of self-management interventions						
					Outcome measure	Theoretical base ^a	Domain of self-management ^b	Formats	Elements	Interventionists	Setting
Studies:	Country	Diagnosis	N	Methodological Quality	Control						
					Usual care	Quality of life	NA	MM	Individual	Family therapy	Clinic
						Symptoms			Education		
Laffel et al. 2003 [65]	USA	Diabetes	N = 100 I = 50 C = 50	Moderate	Usual care				Goal-setting skills	Research assistant	
Mcperson et al. 2006 [66]	United Kingdom	Asthma	N = 101 I = 51 C = 50	Moderate	Another specific intervention	Disease knowledge	NA	MM	Individual	Education	Home
						Symptoms			Self-monitoring		
									Problem-solving skills		
Mulvaney et al. 2010 [67]	USA	Diabetes type 1	N = 49 I = 31 C = 18	Moderate	Usual care	Symptoms	CSL	MM	Individual	Problem-solving skills	Online
						Adherence	Self-determination theory		Peer-support		
Naar-King et al. 2013 [68]	USA	HIV	T = 76 I = 36 C = 40	High	Another intervention	Adherence	Principles of motivational interviewing	MM	Individual	Education	Home
						Symptoms			Goal setting skills		
									Telemedicine		

Background		Participants		Study characteristics		Components of self-management interventions						
Studies:	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventionists	Setting
Newcombe et al. 2012 [69]	Australia	Chronic respiratory condition	N = 39 I = 19 C = 20	High	Waitlist	Psychological outcomes	NA	MM	Individual	Education	NA	Online
						Dealing with the chronic condition in daily life			Group (online)	Self-monitoring		
										Peer-support		
Ng et al. 2008 [70]	China	Asthma	N = 37 I = 20 C = 17	Moderate	Waitlist	Quality of life	NA	MM+RM+EM	Group	Education	NA	Clinic
						Symptoms				Family therapy		
Nunn et al. 2006 [71]	Australia	Diabetes type 1	N = 139 I = 60 C = 63	Moderate	Usual Care	Disease knowledge	NA	MM	Individual	Education	NA	Home
										Tele medicine		
										Self-monitoring		
Palermo et al. 2009 [72]	USA	Chronic pain	N = 48 I = 26 C = 22	High	Waitlist	Symptoms	CBT, CSL	MM	Individual	Education	Psychologist	Online
						Psychological outcomes				Cognitive restructuring		
										Relaxation training		

Background	Participants			Study characteristics			Components of self-management interventions					
Studies:	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventionists	Setting
Pulgaron et al. 2010 [73]	USA	Asthma	N = 41 I = 20 C = 21	Moderate	Usual care	Disease knowledge Dealing with the chronic condition in daily life	NA	MM	Group	Education Problem solving skills Peer-support	Psychologist (under-graduate degree)	Camp
Rhee et al. 2011 [74]	USA	Asthma	N = 91 I = 46 C = 45	Moderate	Another specific intervention	Quality of life Symptoms	NA	MM+RM	Group	Discussion Education Tele-medicine Peer support	NA	Camp
Scholten et al. 2013 [75]	Netherlands	Chronic condition	N = 194 I1 = 71 I2 = 49 C = 74	Moderate	Waitlist	Psychological outcomes	NA	MM + EM + RM	Group	Education Relaxation techniques Cognitive restructuring Social skills Family therapy	Psychologist	Clinic School

Background		Participants		Study characteristics		Components of self-management interventions						
Studies:	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventionists	Setting
Shames et al. 2004 [76]	USA	Asthma	N = 119 I = 59 C = 60	Moderate	Usual care	Symptoms	NA	MM	Individual	Education	Case manager Clinician	Clinic Home
						Disease knowledge				Tele-medicine	Nurse	
Staab et al. 2006 [77]	Ger-many	Atopic derma-titis	N = 823 I = 446 C = 377	Moderate	Another specific inter-vention	Symptoms	NA	MM+EM	Group	Education	Derma-tologist Pediatrician Psychologist Dieticians	Clinic
										Peer-support Relaxation training		

Background	Participants		Study characteristics		Components of self-management interventions							
Studies:	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventionists	Setting
Stinson et al. 2010 [78]	Canada	Juvenile Idiopathic Arthritis	N = 46 I = 22 C = 24	High	Another specific intervention	Quality of life Symptoms Psychological outcomes Adherence Disease knowledge Dealing with the chronic condition in daily life	NA	MM+RM+EM	Individual	Education Tele-medicine	Psychologist (undergraduate degree)	Online
Stulemeijer et al. 2005 [79]	Netherlands	Chronic Fatigue Syndrome	N = 71 I = 36 C = 35	High	Waiting list	Symptoms School attendance	CBT	MM+RM+EM	Individual	Education Cognitive restructuring	Psychologist	Clinic
Velsor-Friedrich et al. 2005 [80]	USA	Asthma	N = 52 I = 28 C = 24	Low	Usual care	Symptoms	Orem's Self-Care Deficit Theory of Nursing	MM+RM	Individual Group	Education Peer-support	Nurse practitioner	School

Background		Participants		Study characteristics			Components of self-management interventions						
Studies:	Country	Diagnosis	N	Methodological Quality		Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventionists	Setting
Walders et al. 2006 [81]	USA	Asthma	N = 175 I = 89 C = 86	Moderate	Usual care		Symptoms	NA	MM	Individual	Education	Nurse	Clinic
							Quality of life				Problem-solving skills	Asthma social worker	
											Telemedicine	Psychologist	
Wiecha et al. 2005 [82]	USA	Asthma	N = 58 I = 37 C = 20	Low	Usual Care		Symptoms	SCL	MM	Individual	Education	Physician	Home
							Adherence				Self-monitoring	Research assistant	
											Peer-support		
											Discussion		

Background		Participants		Study characteristics		Components of self-management interventions						
Studies	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventionists	Setting
Wysocki et al. 2007 [83]	USA	Diabetes	N = 104 I = 28 C1 = 31 C2 = 26	Moderate	Another specific intervention Usual care	Symptoms Adherence	NA	MM+ RM	Individual	Problem solving skills Family therapy Communication skills Education Cognitive restructuring	Psychologist	Clinic

^a NA= Not available; CSL = Cognitive social learning theory; CBT = cognitive behavioral theory

^b MM = Medical Management; EM = Emotion Management; RM = Role Management (according Lorig & Holman, 2003)

4

Social participation and psychosocial outcomes of young adults with chronic physical conditions: Comparing recipients and non-recipients of disability benefits

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ABSTRACT

Background: Little is known about any differences between young people with chronic physical conditions who do and do not apply for disability benefits in young adulthood for providing insights for future policy and rehabilitation care.

Objective: We aimed to identify predictors during adolescence of receiving disability benefits in young adulthood and to compare recipients and non-recipients of benefits in social participation and psychosocial outcomes in young adulthood.

Methods: Follow-up study of 18 to 25 year olds with various chronic conditions who at adolescent age completed a web-based survey (n=518; T0). The outcome was receiving disability benefits (yes or no). Associations with background characteristics, social participation, and impact of the chronic condition were explored with stepwise multivariate modelling, using T0 variables. Differences between recipients and non-recipients were explored using chi-square tests and t-tests.

Results: Receiving disability benefits in young adulthood was associated with greater extent of physical disability, receiving less special education, absenteeism at school/work, and low health-related quality of life during adolescence. In young adulthood, recipients of benefits reported higher perceived impact of the chronic condition on their school/work career and lower quality of life than non-recipients. Social participation varied across domains.

Conclusion: This study provides important insights into the characteristics of a vulnerable subgroup of young people with chronic physical conditions. Disability benefit recipients experienced more impact of their chronic condition and reported a lower health-related quality of life over time than non-recipients. Rehabilitation professionals are encouraged to use patient-reported outcomes to address the lived experiences and screen the need for psychosocial support of this vulnerable subgroup of young people with chronic physical conditions.

INTRODUCTION

Employment fosters the acquirement of life skills and autonomy needed to participate in society [1]. It increases people's sense of control over their own lives, helps develop their self-identity, and positively influences income levels and occupational achievements during the lifespan [1, 2]. Therefore, starting a work career is an important developmental milestone for young adults. However, finding and maintaining employment is a major challenge for young adults with chronic physical conditions [3, 4]. As compared with healthy peers, non-healthy young adults show lower work participation rate [4-6]. Reported employment rates vary, but rates of about 30% are common in both Europe and the United States [7, 8]. In The Netherlands, 39% to 45% of young adults with chronic physical conditions are employed [8, 9] versus 64% to 72% of their healthy peers [7, 9]. Reported barriers to employment include limited accessibility of work places, discrimination, inadequate possibilities for transportation, and lack of social support [3, 4, 10, 11]. Unemployment in turn negatively affects the financial situation, psychological well-being, social interactions, and quality of life of such people [3, 4].

Rehabilitation healthcare teams have taken steps to foster the work participation of young adults with chronic physical conditions [12-14]. Also, on the macro level, policies aimed at improving their work participation and quality of life have been implemented. In The Netherlands, young adults who are partially or fully incapable of working due to childhood onset of a chronic condition may be entitled to a benefit under the scheme for young disabled persons: Wajong Act (the Disablement Assistance Act for Handicapped Young Persons) [15]. People may apply for Wajong if they are at least 25% occupationally disabled before their 17th birthday or became occupationally disabled during their education. The disability benefit consists of (supplementary) income support, and those who are fit to work are supported in finding and sustaining employment [15, 16].

The Netherlands National Employee Insurance Agency has assessed young adults' work ability and capacity. The number of benefit recipients rose from 147000 in 2005 to 249000 in 2015 [15, 17]. About 25% of the recipients of benefits were employed, whereas 60% of all recipients were found fit to work [18]. In other countries, the number of disability benefit claims has risen in recent years, with lower employment rates reported for benefit recipients [8].

Recent studies of young adults receiving disability benefits emphasized that they had a less favourable developmental trajectory while growing up as compared with healthy age-mates [16, 19, 20]. In young adulthood, they report lower health-related quality of life (HRQoL) and are at increased risk for anxiety and depression [19, 21]. Therefore, psy-

chosocial support during childhood and adolescence is recommended [20]. Still, little is known about any differences between young people with chronic physical conditions who apply for disability benefits and those who do not [21].

This study explored the association between characteristics during adolescence and the likelihood of receiving disability benefits in young adulthood among a cohort of young people with chronic physical conditions. In addition, we studied the differences between recipients and non-recipients of benefits on participation and psychosocial outcomes in young adulthood. Our aim was to provide insights for future policy and rehabilitation care.

MATERIAL AND METHODS

Study design and participants

A longitudinal survey study was conducted whereby participants of a Web-based survey in 2006 (T0) were re-invited for a similar survey six years later (T1) [22]. The Web-based follow-up questionnaire was basically the same as the previous questionnaire, except that questions on social participation and source of income were added. It was pilot-tested in face-to-face (n=7) and telephone (n=3) interviews with young university students with a chronic physical condition who were not included in the final sample. From the pilot test, some questions were rephrased or deleted to reduce the response time.

In 2006, participants were adolescents aged 12 to 18 years who had been under treatment at the Erasmus MC–Sophia’s Children’s Hospital Rotterdam for more than 3 years. They had a variety of chronic physical conditions. Those with intellectual disabilities were excluded. In 2012, contact addresses and death notices were retrieved from the hospital’s electronic patient records. Eligible young adults were sent a patient information letter and a unique password to log into a secured website. Included was a postcard on which they could state they did not want to participate. Patients who did not respond within one month were sent a reminder by mail. After another month, non-responders were reminded by a phone call. Respondents were entered in a lottery to win one of 25 cookbooks, two smart phones, or an iPad provided by local suppliers. The Erasmus MC medical ethical review board approved the study (MEC 2012-022) and all data were processed anonymously. Participants electronically provided consent for their participation in the study.

Measures and study outcomes

Main outcome

At T1, respondents were asked if they were recipients of benefits within the framework of the Wajong Act (yes, no) [16].

Independent variables

Age, medical diagnosis (classified according to International Statistical Classification of Diseases and Related Health Problems [ICD-10]), educational level [low or medium (primary school, secondary general vocation or lower vocational education); high (higher educational institutions or university)], and type of education followed in the past (mainstream, special education) were recorded at both T0 and T1; data from the T0 questionnaire established gender (female, male) and age at onset of the chronic condition (congenital and 1-5, 6-12, >12 years). The extent of physical disability had been measured at T0 with 10 items scored on a 4-point Likert scale (1 = no physical limitations; 2 = some difficulty; 3 = great difficulty; 4 = I cannot do it) [23]. A total score was computed by summing the scores (Cronbach's $\alpha = 0.91$) [23]. At T0, data on medical diagnosis and onset of the chronic condition was verified in patient medical records.

Self-management, as reflected by disease-related self-efficacy, was measured at T0 with the On Your Own Feet Self-Efficacy Scale (OYOF-SES) by a 4-point Likert scale (from 1 = no, definitely not, to 4 = yes, certainly) [24]. The OYOF-SES consists of three domains: coping with the condition (four items, Cronbach's $\alpha = 0.82$), knowledge about the condition (six items, Cronbach's $\alpha = 0.78$) and competencies during consultations (six items, Cronbach's $\alpha = 0.85$).

Perceived impact of the condition was measured at T0 in different ways. Adolescents reported the visibility of their chronic physical condition as well as absenteeism at school or work due to their condition by using a 3-point Likert scale (1 = never; 2 = sometimes; 3 = frequently to always). The experienced burden of visibility of the chronic physical condition was measured by 2 items scored on a 5-point Likert scale (1 = no burden at all; 2 = no burden; 3 = little burden; 4 = burden; 5 = much burden). These two items were summed (Cronbach's $\alpha = 0.80$). The self-reporting short-form DISABKIDS questionnaire (DCGM-10) was used to assess HRQoL at T0 (Cronbach's $\alpha = 0.80$) [25].

Outcome measures at T1

Level of autonomy in social participation was classified by using the Rotterdam Transition Profile (RTP) [26]. The RTP describes participation in seven life areas defined in the International Classification of Functioning, Disability, and Health: finances, employment and education, housing, intimate relationships, sexuality, transportation, and leisure. It

distinguishes four transition phases (0-3). Young people in phases 0 and 1 are still fully dependent on adults (e.g., parents) or display typical child behaviour. Young people in phase 2 experiment with or orient to adult behaviour. Phase 3 refers to full autonomy in participation. Because we were interested in a successful transition to adulthood, we dichotomised the phases as follows: 0 = phases 0-2; 1 = phase 3 [22].

Perceived impact of the chronic condition on current education/vocational participation was measured at T1 with a newly constructed five-item scale. The items were derived from the relevant literature and from interviews with young adults with chronic physical conditions and are reported elsewhere [22]. The impact was rated on a 5-point Likert scale (1 = totally disagree; 2 = disagree; 3 = neutral, 4 = agree; 5 = totally agree). The items were summed (Cronbach's $\alpha = 0.88$).

HRQoL was assessed at T1 by using the self-reporting DISABKIDS questionnaire (DCGM-37) [27], which consists of five domains: independence (6 items, Cronbach's $\alpha = 0.86$); physical (6 items, Cronbach's $\alpha = 0.84$); emotion (7 items, Cronbach's $\alpha = 0.91$); social exclusion (6 items, Cronbach's $\alpha = 0.81$); and social inclusion (6 items, Cronbach's $\alpha = 0.83$). Responses are given on a 5-point Likert scale (1 = often; 2 = quite often; 3 = sometimes; 4 = almost never; 5 = never). A general score was computed by combining all items (Cronbach's $\alpha = 0.95$). HRQoL scores were transformed to a 0–100 scale.

Statistical analysis

Backward logistic analysis was used to detect selective response; determinants of study non-response were evaluated by odds ratios (ORs) with 95% confidence intervals (95% CI), and Nagelkerke R^2 indicated the proportion of explained variance. Model fit was tested with the Hosmer–Lemeshow test. The study sample was described with descriptive statistics. Differences between background characteristics of benefit recipients and non-recipients were tested by Student t test (continuous variables) and chi-square test (nominal variables). Associations between the likelihood of a disability benefit claim and other variables (Cramer's V and Phi for ordinal or nominal variables; and Pearson's r for interval variables) were established to determine which variables should be included in the regression analyses. Only variables significantly correlated ($p < 0.05$) with the outcome variable were included in the regression model. Stepwise multivariable linear regression was used to identify factors associated with a disability benefit claim, starting with background variables, then the T0 variables. Multicollinearity was checked by calculating the Variance Inflation Factor (VIF). Chi-square test was used to study differences between benefit recipients and non-recipients in autonomy in social participation (T1). Finally, independent t tests were used to detect differences between recipients and non-recipients in HRQoL and perceived impact of the chronic physical condition on

school and work career (T1). Data were analysed with SPSS 20. $P < 0.05$ was considered statistically significant.

RESULTS

Among the 1039 participants in the original study, 13 had died and 25 could not be traced. Consequently, 1001 were invited to participate in this study, 88 of whom returned a postcard stating that they declined to participate. Eventually, 518 young adults (net response: 51.8%) submitted the survey. Backward logistic regression analysis showed that non-response was associated with male gender (OR 0.57; 95% CI 0.43–0.74) and non-Dutch ethnicity (0.49; 0.33–0.74) ($\chi^2 = 29.0$, degrees of freedom = 2; $p < 0.001$; $R^2 = 0.04$; Hosmer–Lemeshow test, $p = 0.773$). Six respondents (1.2%) did not report whether they received social security benefits and were excluded from the analysis. Hence, the final sample numbered 512, of which 101 respondents received a disability benefit. The characteristics of the study sample are presented in Table 1. Congenital malformations, deformations and chromosomal abnormalities were most frequent, followed by neoplasms; endocrine, nutritional, and metabolic diseases; diseases of the eye and adnexa and the ear and mastoid process; diseases of the musculoskeletal system and connective tissue; and diseases of the digestive system.

Association with receiving a disability benefit

To determine which variables should be included in the regression analyses, associations were tested (Table 2). The multivariate model for receiving a disability benefit explained 44% of the total variance ($\chi^2 = 307.44$, degrees of freedom = 10; $p < .001$; Hosmer–Lemeshow test, $p = .54$) (Table 3). VIF scores ranged from 1 to 2, indicating absence of multicollinearity. Receiving a disability benefit in young adulthood was associated with the extent of physical disability (OR 1.28; 95% CI 1.10–1.48) and less special education during adolescence (0.15; 0.07–0.33) as well as absence from school or work (1.54; 1.10–2.10) and low HRQoL during adolescence (0.97; 0.95–0.99).

Social participation

We found no significant differences between benefit recipients and non-recipients in achieved autonomy in the life areas employment ($p = 1.00$), social relationships ($p = 0.10$), and housing ($p = 0.78$) (Table 4). As compared with non-recipients, recipients were less often involved in intimate and sexual relationships (34.7% vs 57.1%; $p < 0.001$) and less often independently organized transportation (75.8% vs 96.4%, $p < 0.001$) and attended social events (leisure) (33.7% vs 59.9%; $p < 0.001$). As expected, benefit recipients were more often financially self-supporting than were non-recipients (100% vs 29.0%; $p < 0.001$).

Table 1. Description of the study sample for recipients and non-recipients of disability benefits

	No. Total study sample		Benefit recipients n=101		Benefit non-recipients n=411		p-value**
	No.	Total study sample	No.	Benefit recipients n=101	No.	Benefit non-recipients n=411	
Age T1 [18-25], mean (SD)	508	20.25 (3.3)	101	20.50 (4.04)	407	20.19 (3.10)	0.41
Gender, male, no. (%)	512	196 (38.3)	101	30 (29.7)	411	166 (40.4)	0.03
Disease, no. (%)	510						
Congenital malformations, deformations and chromosomal abnormalities		146 (28.6)	101	28 (27.9)	409	118 (28.9)	0.46
Neoplasms		79 (15.5)		8 (7.9)		71 (17.4)	0.01
Endocrine, nutritional and metabolic diseases		66 (12.9)		17 (16.8)		49 (12.0)	0.13
Diseases of the eye and adnexa, and the ear and mastoid process		64 (12.5)		27 (26.7)		37 (9.0)	<0.001
Diseases of the musculoskeletal system and connective tissue		46 (9.0)		6 (5.9)		40 (9.8)	0.16
Diseases of the digestive system		28 (5.5)		5 (5.0)		23 (5.6)	0.51
Other		81 (15.9)		10 (9.9)		71 (17.4)	<0.05
Extent of physical disability T0 [range 10-40]*, no. (%)	509	33 (11.17)	99	14.36 (6.67)	410	10.40 (1.11)	<0.001
Age of onset of chronic condition T0, years, no. (%)	512		101		411		
Congenital		290 (56.6)		68 (76.3)		222 (54.0)	0.01
0-5		64 (12.5)		9 (8.9)		55 (13.4)	0.15
6-12		120 (23.4)		19 (18.8)		101 (24.6)	0.14
>12		38 (7.4)		5 (5.0)		33 (8.0)	0.20
Educational level T1, low or medium, no. (%)	500	271 (54.2)	95	63 (66.3)	405	208 (51.4)	0.006
Special education T1, yes, no. (%)	508	88 (17.2)	101	55 (54.5)	407	33 (8.1)	<0.001

* Theoretical range

** Comparing recipients and non-recipients by paired-sample t test (continuous variables) or chi-square test (nominal variables)
T0, participants of a Web-based survey in 2006; T1, participants re-invited for a similar survey 6 years later

Table 2. Associations between variables (at T0) and a disability benefit claim (at T1)

	No.	Cramers's V, Phi, or Pearson's correlation	<i>p</i> -value
Background characteristics			
Gender	512	-0.08	0.09
Disease			
Congenital malformations and chromosomal abnormalities	146	-0.10	0.82
Neoplasms	79	0.10	0.02
Endocrine, nutritional and metabolic diseases	66	0.06	0.19
Diseases of the eye and adnexa, and the ear and mastoid process	64	0.21	<0.001
Diseases of the musculoskeletal system	46	-0.05	0.23
Diseases of the digestive system	28	-0.01	0.79
Extent of physical disability			
Onset chronic condition	512	0.11	0.11
(congenital)	512	0.11	0.02
Educational level	475	-0.15	<0.001
Special education	477	0.44	<0.001
Self- management			
Self-efficacy			
Total	480	-0.03	0.57
Coping	491	-0.00	0.94
Knowledge	484	-0.03	0.48
Competence	481	-0.01	0.87
Perceived impact of the condition			
Visibility of the chronic condition	511	0.32	<0.001
Absence from school or work	505	0.18	<0.001
Experienced burden	511	0.16	<0.001
Health-related quality of life(DISABKIDS-10)	506	-0.28	<0.001

Table 3. Factors associated with receiving disability benefits (n=512)

	Step 1			Step 2		
	Beta (SE)	OR (95% CI)	p-value	Beta (SE)	OR (95% CI)	p
Background characteristics						
Extent of physical disability	0.35 (0.08)	1.43 (1.22-1.66)	<0.001	0.25 (0.07)	1.28 (1.10-1.48)	<0.001
Educational level	0.33 (0.32)	1.39 (0.75-2.59)	0.30	0.20 (0.33)	0.22 (0.64-2.32)	0.55
Special education	-2.04 (0.39)	0.13 (0.06-0.28)	<0.001	-1.92 (0.41)	0.15 (0.07-0.32)	<0.001
Disease						
Neoplasms	-0.33 (0.47)	0.72 (0.29-1.81)	0.48	-0.57(0.50)	0.57 (0.21-1.50)	0.25
Diseases of the eye and adnexa, and the ear and mastoid process	0.23 (0.45)	1.26 (0.53-3.02)	0.60	0.25 (0.44)	1.28 (0.54-3.03)	0.57
Onset of chronic condition	0.31 (0.31)	1.37 (0.74-2.53)	0.32	0.26 (0.33)	1.30 (0.67-2.50)	0.44
Impact of the chronic condition						
Visibility of the chronic condition				0.12 (0.08)	1.13 (0.96-1.34)	0.15
Experienced burden				-0.12 (0.09)	0.89 (0.75-1.06)	0.20
Absence from school or work				0.43 (0.17)	1.54 (1.10-2.10)	0.01
Health-related quality of life (DISABKIDS-10)				-0.03 (0.01)	0.97 (0.95-0.99)	0.002
Explained variance of the final model	Nagelkerke R ² = 0.39			Nagelkerke R ² = 0.44		
Model	$\chi^2(6) = 328.34, p<0.001$			$\chi^2(10) =307.44, p<0.001$		

Psychosocial outcomes

Recipients reported a higher perceived impact of the chronic physical condition on their school and work career than did non-recipients (15.12 ±5.13 vs 8.89 ±4.60, *p*<0.001) (Table 4). Also, they experienced a lower general HRQoL (65.92±17.38 vs 80.86 ±14.15, *p*<0.001). HRQoL was also lower on each of the subdomains.

DISCUSSION

For a cohort of young people with chronic physical conditions, we explored predictors during adolescence of receiving disability benefits in young adulthood and compared social participation and psychosocial outcomes in young adulthood for recipients and non-recipients of benefits. Receiving a disability benefit later in life was associated with greater extent of physical disability, receiving less special education during adolescence, increased perceived impact of the chronic physical condition on their school career, and reduced HRQoL during adolescence. Recipients of benefits perceived a higher impact of their chronic physical condition on their school career and reported a lower HRQoL than non-recipients. Also, they lagged behind in terms of having an intimate relationship and reported less autonomy in transportation and leisure activities.

Table 4. Differences between disability benefit recipients and non-recipients in autonomy in social participation (T1) and psychological outcomes (T1)

	Benefit recipients (n=101)	Benefit non-recipients (n=411)	χ^2 / t	p-value*
Social participation, n (%)				
Finances	101 (100.0)	118 (29.0)	166.36	<0.001
Employment	27 (26.7)	107 (26.3)	.01	1.00
Housing	21 (20.8)	78 (19.2)	.14	0.78
Social relationship	31 (32.6)	163 (42.1)	2.86	0.10
Intimate relationship	33 (34.7)	221 (57.1)	15.31	<0.001
Transportation	72 (75.8)	373 (96.4)	45.64	<0.001
Leisure	32 (33.7)	232 (59.9)	21.24	<0.001
Perceived impact of the chronic condition on school/work career, mean (SD)	15.12 (5.13)	8.89 (4.60)	-11.89	<0.001
Quality of life, mean (SD)				
General score	65.92 (17.38)	80.86 (14.15)	7.77	<0.001
Physical domain	55.00 (21.52)	74.80 (21.07)	8.18	<0.001
Emotion domain	65.07 (23.70)	78.65 (19.77)	5.76	<0.001
Independence domain	72.28 (19.02)	86.49 (14.12)	6.83	<0.001
Social exclusion domain	69.87 (19.45)	86.36 (16.40)	7.62	<0.001
Social inclusion domain	64.47 (21.50)	79.08 (17.99)	6.11	<0.001

* P<0.05 comparing recipients and non-recipients by paired sample t-test (continuous variables) and chi-square tests (nominal variables)

Our study adds to the literature by defining and characterising a subgroup of young people with chronic physical conditions even more vulnerable with a delay in psychosocial development and as such underlines the need for a lifespan perspective on care for this group. Roebroek and colleagues [28] earlier acknowledged the need for incorporating a lifespan perspective throughout pediatric, transition, and adult rehabilitation care. For instance, in England, deployment of Young Adult Teams of health care professionals was useful in supporting young adults' social participation [29]. Other options introduced in The Netherlands are group-based interventions aimed at single or multiple participation domains, or discussing patient-reported outcomes during consultations [12, 30, 31]. The importance of psychosocial support for adolescents and young adults with a chronic physical condition was previously described [19-21, 32]. Still, these days, transitional care still mostly focuses on medical management issues and less attention is paid to emotional and social aspects of having a chronic condition [33]. Yet, identifying and paying attention to adolescents' lived experiences and needs in the areas of social participation during adolescence is essential for them to achieve their full potential and a satisfying adult life [34]. Research showed that rates of participation differ from healthy peers [22] and also social participation can be less diverse in young people with a chronic condition (e.g., less social or skill-building activities [35]). Therefore, patient-reported outcomes should be monitored on a regular basis. To this end, specific tools are available for psychosocial screening, encouraging independent

behaviors (through the use of individual transition plans such as the Skills for Growing Up tool [30]), and monitoring HRQoL [31] during medical consultations in paediatric and adult rehabilitation care.

Recipients of benefits often had less special education during adolescence, were often absent from school or work during adolescence and experienced more barriers in school/work participation in young adulthood, which underlines the need to pay attention to and provide support for school and work participation [3, 36]. Being engaged in mainstream education and doing temporal or seasonal work during adolescence is important for youth with disabilities because it could provide a context for developing life skills and exploring interests and abilities [11, 36]. Such life skills include “psychosocial abilities for adaptive and positive behaviour that enable individuals to deal effectively with the demands and challenges of everyday life” [37]. At schools for mainstream education, adolescents may receive less support from their teachers, friends, and service providers, which could hinder their opportunities to develop these skills and to fully participate in school [38]. Thus, being encouraged to develop independent life skills may also be essential, especially for adolescents in mainstream education settings.

Young people with a childhood onset of chronic condition must make a well-considered decision on the application for disability benefit. A need for financial compensation and support at finding and maintaining employment could be reasons for applying. However, being a benefit applicant could also negatively affect employment opportunities, for example, by reinforcing an employers’ prejudice. Therefore, rehabilitation professionals should discuss these advantages and disadvantages with young people and their parents.

Recipients of benefits who are fit to work could get support in finding and sustaining employment. Still, we found that employment rates did not differ between recipients and non-recipients of benefits. A low work ability among recipients of benefits could be an explanation for this finding. However, we had no information on the work ability of our respondents and were therefore not able to distinguish between benefit recipients who were (partially) able to work or those not able to work.

We found an association between the extent of physical disability and the likelihood of a benefit claim, which deserves further investigation. To unravel differences in social participation and psychosocial outcomes, subgroups of benefit recipients should be studied. The employment rates of recipients and non-recipients of benefits are still lower than those of healthy age-mates (63.3%) [39], which indicates the need for support in finding and maintaining employment for young people with chronic physical

conditions. Recently, Dutch legalisations aimed at improving the work participation among young people with chronic physical conditions were amended. Young people with chronic physical conditions who do not have any work ability can receive disability payments. In contrast, young people with chronic physical conditions who are (partially) able to work, should be supported by municipalities to find competitive employment. However, resources for providing support are reduced these days. Since disability benefit recipients are a vulnerable group, support for this group should be priority.

We also found that recipients of benefits were more often financially self-supporting than were non-recipients (100% vs. 29%). This suggests that the benefits are substantial and could in fact be an inducement for young people with chronic physical conditions to apply for these benefits. Still, people could be motivated for a job for other reasons than income (e.g., psychological well-being, social interactions, and self-development [3, 4]). Therefore, financial support does not necessarily have a negative influence on work participation.

Our study included a large sample of young people with a variety of chronic physical conditions. The sample was heterogeneous in terms of congenital and acquired conditions. Yet with this heterogeneity, we could not explore differences between recipients and non-recipients of benefits within all included diagnosis groups. However, we analysed associations between receiving disability benefits and the five most common diagnoses and found no significant associations. In addition, studying chronic physical conditions in general could be considered a strength, because all young people growing up with a chronic physical condition face the similar adaptive challenges.

The survey non-response was substantial (48.2%) especially among males and young adults of non-Dutch ethnicity. This response may have affected the outcomes, in that previous research showed male gender as a prognostic factor for work participation [40]. Also, we found gender related to a disability benefit claim. Finally, at T0 we administered only the short-version of the HRQoL questionnaire. Therefore, we could not investigate associations between quality-of-life domains and the likelihood of receiving disability benefits.

Only personal or internal factors were included in our predictive model. However, the likelihood of a disability benefit claim also depends on social or external factors such as national employment options. We can only conclude that providing psychosocial support during childhood may decrease the likelihood of a claim. However, more research is needed to investigate the influence of external factors.

CONCLUSION

This study provides important insights into the development of characteristics of a vulnerable subgroup of young people with chronic physical conditions over time. Claiming a disability benefit in young adulthood was associated with reporting a greater extent of physical disability, less special education, greater impact of the condition on school functioning, and low HRQoL. Moreover, the impact adolescents experienced on vocational functioning and the low HRQoL persisted over time. In addition, in young adulthood, recipients of benefits lagged behind in social participation on several domains. Rehabilitation professionals should be aware of this and could use various tools to address and screen lived experiences and the need for support for participation and psychosocial development of this vulnerable subgroup of young people with chronic physical conditions.

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CONFLICT OF INTEREST

The authors declare no conflicts of interest.

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5

Effectiveness and characteristics of interventions to improve work participation in adults with chronic physical conditions: a systematic review

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ABSTRACT

Purpose: Chronic physical conditions often negatively affect work participation. The objective of this systematic review is to investigate the effectiveness and characteristics of vocational rehabilitation interventions for people with a chronic physical condition.

Methods: Searches in five databases up to April 2020 identified 30 studies meeting our inclusion criteria. Two reviewers independently assessed and extracted data. The Grading of Recommendation, Assessment, Development and Evaluation (GRADE) framework was used to evaluate quality of evidence for three outcome measures related to work participation.

Results: All vocational rehabilitation interventions consisted of multiple components, but their characteristics varied widely. Analysis of 22 trials yielded a moderate positive effect with moderate certainty of interventions on work status; analysis of five trials with low risk of bias showed a large positive effect with moderate certainty (risk ratio 1.33 and 1.57 respectively). In addition, in eight studies we found a moderate to small positive effect with low certainty on work attitude (standardized mean difference=0.59 or 0.38 respectively). We found no effect on work productivity in nine studies.

Conclusion: The systematic review of the literature showed positive effects of vocational rehabilitation interventions on work status and on work attitude; we found no effect on work productivity.

INTRODUCTION

Chronic physical conditions often result in disability, such as pain and physical limitations, and restrictions in daily activities and participation in society. Participation restrictions frequently include temporary or permanent health-related problems in work participation, such as (involuntary) part-time employment, difficulty meeting work demands (for example: work hours, work pace), difficulty in performing work tasks, balancing work and home life, as well as dealing with sick leave, and job loss or unemployment [1-6]. Due to the increasing number of people with a chronic physical condition and their problems with employment, their work participation is a growing concern in society [1, 6].

About 30% of employees with a chronic condition experiences problems with employment related to their condition [3]. Despite disease-specific differences, there are generic characteristics that can be considered common consequences of a chronic physical condition that hamper work participation, such as pain, fatigue and functional disabilities, variability of symptoms, an unpredictable course of symptoms, and long-lasting impact of consequences [3, 5-8]. As a result, people with chronic physical conditions may face many similar challenges and adaptive tasks to participate in work [9]. In addition, a systematic review found that most of the factors associated with work participation of workers with a chronic physical condition are independent of the diagnosis [10]. Therefore, the use of a generic approach to improve the work participation of persons with a chronic physical condition might be appropriate. However, an overview of interventions and evidence on the effectiveness of interventions to enhance work participation of people with chronic physical conditions, irrespective of diagnosis, is lacking.

The current systematic review investigates the effectiveness and characteristics of vocational rehabilitation interventions for people with a chronic physical condition. For this study, chronic back pain was excluded because vocational interventions for patients with chronic back pain have been systematically reviewed and published [11-14].

METHODS

A systematic review of the literature was performed: such reviews seek to systematically search for, appraise and synthesize research evidence [15]. This type of review allows to explore the effectiveness and intervention characteristics. The study protocol was not registered and has not previously been published.

Search strategy

A systematic extensive electronic search was conducted in the databases Medline, Cinahl, Cochrane controlled trials register, Embase and PsycINFO up to April 2020. The search strategy was developed based on literature [16, 17], group discussions among the authors, and preliminary searches to inform the strategy. The final search strategy employed variations and Boolean connections (AND, OR, NOT) of MeSH terms, subject headings and keywords related to chronic physical conditions, vocational rehabilitation, and work participation. In addition, we used the search strategy for randomized controlled trials and controlled clinical trials as recommended by the Cochrane collaboration [18]. The search strategy was adapted according to the particular database, using database thesauruses to identify relevant variations of these terms. The search strategy used in Medline is presented in Box I. In addition, the reference lists of the identified original papers were checked for additional relevant studies.

Box I Search strategy (Medline)

((((Clinical trial[pt] OR randomized[tiab] OR placebo[tiab] OR clinical trials[mh] OR randomly[tiab] OR trial[ti]) NOT (animals[mh]))) AND (program* OR training OR education OR rehabilitation OR multidisciplinary OR therapy)) AND ("Work"[Mesh] OR "Employment"[Mesh] OR "Employment, Supported"[Mesh] OR "Occupations"[Mesh] OR job OR employment OR vocation)) AND (("Chronic Disease"[Mesh] OR "Disabled Persons"[Mesh] OR chronic OR disability or disabilities) NOT (mental or mentally or psychiatric))

Study selection

Retrieved records (n=13156) were imported into Endnote and duplicates were removed (n=3465). Inclusion criteria for this review were: (i) Population: working-age adults (18-65 years) with a chronic physical condition, other than chronic back pain, lasting ≥ 3 months or that can be categorised as long-lasting based on disease characteristics (for example: rheumatoid arthritis) (ii) Intervention: studies focusing on vocational rehabilitation interventions containing specific elements to improve work participation (excluding surgery, medication) (iii) Comparison: no vocational intervention (usual care, waiting list) (iv) Outcome: work participation (v) original controlled trials in the English language and peer-reviewed.

Based upon title and abstract, three authors (JV, MB, PR) independently screened the articles for eligibility and excluded the studies that clearly did not meet one or more inclusion criteria (n=9429). Full-text copies of all other articles (n=119) were obtained and two authors (JV, MB) independently decided on the inclusion of these studies, based upon full text. At all stages of selection, disagreements in the independent decisions were resolved by discussion until consensus was reached. If both reviewers did not reach consensus, a third author was consulted (HM). The selection process and reasons for exclusion are shown in Figure 1.

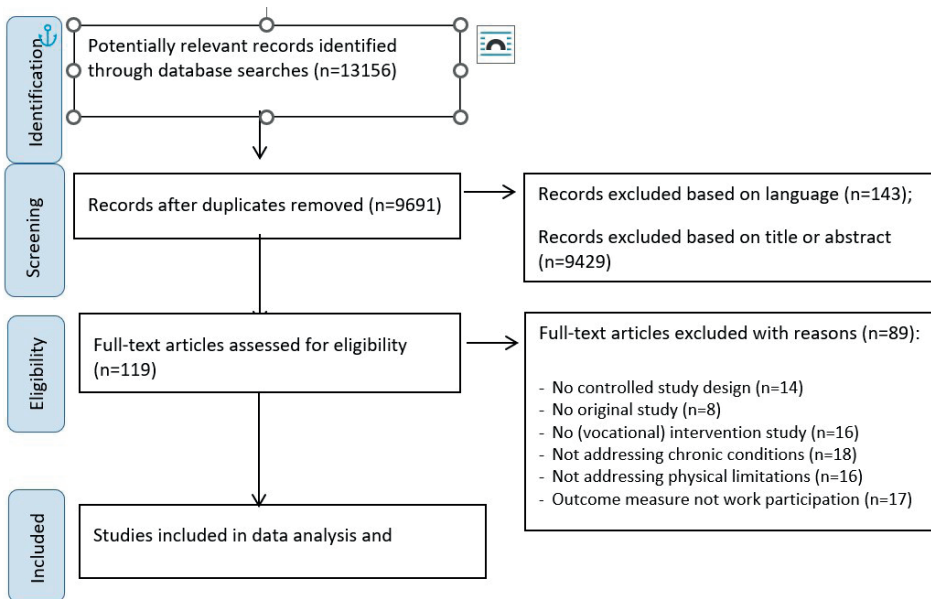


Figure 1 Flowchart of identification and selection of studies

Data extraction

Two reviewers (JV, MB) extracted data on study design, study sample, characteristics and content of intervention [format (individual, group, combined), approach and professionals (mono- or multidisciplinary) duration (short, moderate and long) and intensity (low and high), and focus (vocational, cognitive-behavioural or physical)], outcome measures and study results using a pre-designed data extraction form. Duration of interventions was categorized as short (<10 weeks), moderate (10-16 weeks) or long (>16 weeks), based on median (10 weeks) and mean (16 weeks) values of duration of interventions, and intensity as low (≤ 40 hrs in total) or high (>40 hrs), based on mean number of hrs. The focus of interventions was characterized as vocational, cognitive-behavioural or physical, based on the functional domain that was primarily targeted to improve work participation.

Outcome measures were inductively derived from the data, i.e:

1. *Work status*, reported as dichotomous outcome (yes/no) presenting the proportion of the study sample achieving return to work, employment or job maintenance;
2. *Work productivity*, reported as continuous outcome presenting work productivity (hours per week worked) or duration of sick leave;
3. *Work attitude*, reported as continuous outcome presenting scores on work readiness (action to find a job), employment activities, employability or self-efficacy at work.

Two reviewers (JV, MB) independently assigned the study outcomes to these three outcome measures. Other outcomes from the selected studies were not included in this review.

Methodological quality assessment

For each of the 30 included studies, two reviewers (JV and MB) independently assessed the methodological quality using the Effective Public Health Practice Project Quality Assessment Tool (EPHPP) for Quantitative Studies (https://merst.ca/wp-content/uploads/2018/02/quality-assessment-tool_2010.pdf). This tool assesses six domains or potential sources of Risk of Bias (RoB): 1) selection bias; 2) study design; 3) confounders; 4) blinding; 5) data collection method; and 6) withdrawals and dropouts. We decided to use the EPHPP tool rather than the Cochrane Collaboration Risk of Bias Tool, because the EPHPP tool was developed for use with various intervention study designs (not only randomized controlled trials), which we intended to include. The scoring of the EPHPP is based on objective guidelines [19]. Consistent with the EPHPP Quality assessment tool dictionary, each domain was rated as strong, moderate or weak and the methodological quality of the study was rated as strong when no domain was rated as weak, moderate if one domain was rated as weak, and low if two or more domains were rated as weak. Any disagreement about the methodological quality was resolved by discussion until consensus was reached.

Data analysis and synthesis

A mixture of strategies was used for data analysis to accommodate the variety of interventions and studies.

First, the content and characteristics of interventions were described based on data-extraction.

Second, we explored effectiveness of interventions for each outcome measure separately. The estimated effects of all studies for each outcome were first visually displayed in forest plots using Review Manager (software version 5.3) [18, 20]. A random effects model was applied to take into account the possible statistical heterogeneity of the studies. For the dichotomous outcome work status, the risk ratio (RR) was used to compare effect sizes [18, 21]. For the continuous outcomes work productivity and work attitude the standardized mean difference (SMD) was used, since measurement scales differed across studies [21]. Data obtained at 12 months (52 weeks) follow-up were used for the analyses. Otherwise, the duration of follow-up in weeks was added to the study reference (for example: Macedo et al. 2009_26). In the case of missing data, authors were

contacted to request additional information [22]. Studies lacking standard deviations (SD) of mean scores were not included in the forest plots [23].

Third, statistical pooling (meta-analysis) of the outcome measures work status, work productivity and work attitude was performed, for all studies that reported one (or more) of these specific outcome measures, as well as the subset of studies of which the methodological quality was rated as high. Statistical heterogeneity was assessed with Chi^2 and I^2 statistics. The effect size of the dichotomous outcome measure work status was interpreted as high, when the RR was ≤ 0.50 or ≥ 1.50 and as moderate with an RR > 0.50 but ≤ 0.75 or < 1.50 but ≥ 1.25 . Interpretation of the effect size of continuous outcome measures with SMD was based on Cohen's method [24]: small when SMD > 0.20 but < 0.50 , moderate when SMD ≥ 0.50 but < 0.80 and large when SMD ≥ 0.80 .

Finally, we assessed the overall quality of the evidence for each outcome measure using the Grades of Recommendation, Assessment, Development and Evaluation Working Group (GRADE) approach [18, 25-28]. Evidence of randomized controlled trials was rated as high, but could be rated down by one level (into moderate) or by two or three levels (to low or very low quality), with respect to each of five factors: 1) risk of bias: outcome of EPHPP-tool moderate or weak; 2) inconsistency: variability or heterogeneity in magnitude of effect among studies; 3) indirectness of evidence: indirect comparison of interventions within studies; 4) imprecision of results: for dichotomous outcomes full confidence intervals (CI) below 0.75 or above 1.25 or a number of participants of ≥ 300 ; for continuous outcomes full CI below or above the level of a predefined minimal important difference or number of participants ≥ 400 ; 5) likeliness of publication bias: underestimation or overestimation of effect due to selective publication of studies. Publication bias was checked by constructing a funnel plot [29].

RESULTS

Study characteristics

A total of 30 studies met our inclusion criteria for this systematic review [22, 23, 30-57] (see Figure 1). These studies addressed work participation in patients with various chronic physical conditions, such as musculoskeletal disorders and/or musculoskeletal pain ($n=13$), rheumatic diseases ($n=6$), traumatic brain injury ($n=5$), mixed population of various chronic physical diseases ($n=3$), spinal cord injury ($n=2$) and human immunodeficiency virus (HIV) ($n=1$).

Studies were published between 1991 and 2020 and conducted in various countries: the USA (n=10), the Netherlands (n=5), Hong Kong (n=3), Sweden (n=3), Norway (n=2), United Kingdom (n=2), Australia (n=1), Canada (n=1), New Zealand (n=1), South Africa (n=1) and Switzerland (n=1).

Methodological quality of the included studies

The results of the methodological quality assessment are presented in Table 1: 8 studies were rated as high quality, 9 studies as moderate quality and 12 studies as low quality.

Characteristics and content of interventions

An overview of the characteristics of the included studies and interventions is presented in Table 2. With regard to intervention format, 11 of 30 interventions (37%) combined a group program and individual treatment, 13 interventions (43%) only offered individual treatment and 6 interventions (20%) only provided a group program (Table 2 and 3).

Fifteen interventions (50%) applied a monodisciplinary approach and 15 interventions (50%) a multidisciplinary approach. In 10 of the 15 multidisciplinary interventions (67%), the team consisted of healthcare professionals only (for example: physician, physiotherapist, occupational therapist, psychologist); 4 interventions (27%) included also vocational professionals, such as a vocational counsellor or job coach. One team consisted of vocational professionals only. Fourteen interventions (47%) had a short duration, lasting <10 weeks; five (17%) with a high intensity (>40 hrs in total) and nine (30%) with a low intensity. Eight interventions (27%) had a moderate duration of 10-16 weeks, two (7%) with a high and 6 (20%) with a low intensity. Another eight interventions (27%) had a long duration, lasting ≥ 16 weeks, all with a low intensity.

All 30 interventions consisted of multiple treatment components, with a focus on vocational, (cognitive-)behavioural or physical functioning. Interventions primarily targeting vocational functioning (n=16; 53%) to improve work participation, included job searching, job interview skills, specific work training, job placement, vocational counselling, or work place visits. Interventions primarily targeting (cognitive-) behavioural change (n=9; 30%), included coping strategies, problem solving, goal setting, cognitive skills, communication skills, stress management, or empowerment. Interventions targeting physical functioning to improve work participation (n=5; 17%), included exercise training, skills training, graded activity, or work hardening.

Table 1 Methodological quality rating of the included studies using the EPHP tool

Study	Selection bias	Study design (Allocation bias)	Confounders	Blinding (detection bias)	Data collection methods	Withdrawals and drop-outs (attrition bias)	Rating
Hutting et al. 2015 (22)	M	S	S	M	S	M	Strong
Li et al. 2006 (40)	M	S	S	S	S	S	Strong
Li-Tsang et al. 2008 (41)	S	S	S	S	S	S	Strong
Marhold et al. 2001 (43)	S	S	S	M	S	S	Strong
Meyer et al. 2005 (54)	S	S	S	S	M	S	Strong
Ntsiea et al. 2014 (46)	S	S	S	M	S	S	Strong
Sullivan et al. 2006 (50)	M	M	S	M	M	S	Strong
Trexler et al. 2016 (51)	M	S	S	M	S	S	Strong
Allaire et al. 2003 (30)	W	S	S	M	M	M	Moderate
Cheng & Hung 2007 (33)	W	S	S	M	M	S	Moderate
De Buck et al. 2005 (34)	M	S	S	M	W	S	Moderate
Ipsen et al. 2012 (38)	M	S	S	S	S	W	Moderate
Kendall & Thompson 1998 (39)	M	S	W	S	S	M	Moderate
Keysor et al. 2018 (56)	W	S	S	S	S	S	Moderate
Macedo et al. 2009 (42)	M	S	S	W	S	S	Moderate
Salazar et al. 2000 (48)	M	S	S	W	M	S	Moderate
Skouen et al. 2006 (49)	M	S	W	M	S	S	Moderate
Anderson et al. 2007 (31)	W	S	W	M	S	W	Weak
Brattberg et al. 2006 (32)	W	S	S	M	W	S	Weak
Detaille et al. 2013 (35)	W	S	S	M	W	M	Weak
Dorstyn et al. (2019) (57)	W	S	S	M	W	M	Weak
Feuerstein et al. 1993 (36)	M	M	W	M	M	W	Weak
Haffey et al. 1991 (37)	S	M	S	W	W	W	Weak
Hammond et al. 2017 (55)	W	S	W	M	M	M	Weak
Marnetoft & Selander 2000 (44)	M	M	S	M	W	W	Weak
Martin et al. 2012 (23)	W	S	S	M	W	W	Weak
Niemeijer et al. 2010 (45)	W	S	S	W	M	S	Weak
Ottomanelli et al. 2012 (47)	W	S	W	W	W	S	Weak
Van Vilsteren et al. 2016 (53)	W	S	S	W	S	S	Weak
Varekamp et al. 2011 (52)	W	S	M	M	W	M	Weak

S= Strong; M=Moderate; W= Weak.

Rating: Strong (no WEAK rating), Moderate (one WEAK rating), WEAK (two or more WEAK ratings).

Table 2 Characteristics of the included studies and interventions

Study characteristics		Intervention characteristics							
Study	Method	Delivery characteristics			Intervention content				
	Participants (n, diagnosis)	Outcome	Control	Results at 12 months follow-up (unless otherwise noted)	Format	Approach	Duration/ frequency	Focus	Program description
Allaire et al. 2003	N=242; Rheumatic disease	Work status (job loss)	Printed materials only	No significant differences in % of persons with job loss between IG and CG	I	Monodisciplinary	Two 1.5-h sessions within 5 months (total 3 h)	V	Job retention vocational rehabilitation intervention, addressing job accommodation (work barriers and solutions), vocational counselling and guidance.
Anderson et al. 2007	N=52; Chronic pain	Work status (RTW)	Usual care	No significant differences between groups	GI	Monodisciplinary	Multimodal training (UC) followed by 18x training group during 1.5 years (total ± 27 h)	P	Multimodal treatment for 4 weeks (UC) followed by training group based on Psychomotor Physiotherapy (NPMP), emphasizing functional movements, relaxation and incorporating cognitive behavioral therapy.
Brattberg et al. 2006	N=60; Chronic pain and/or burnout	Work status; Work productivity (h/wk worked)	Waiting list	Significantly more persons in IG than in CG had improved work status. Significant group differences in work capacity.	G	Monodisciplinary	20 weeks with weekly sessions (total ± 20 h)	CB	Pain workshop internet course, addressing obstacles to change, expectations and disappointments, self-knowledge, self-destructive behavior, setting limits, stress management, strengths and weaknesses.
Cheng & Hung 2007	N=94; MSD	Work status (RTW)	Usual care	Significant higher RTW rate in IG than in CG after 4 weeks.	I	Monodisciplinary	4 weeks, 3 sessions/week (total ± 12 h)	P	Workplace-based work hardening training. Job-specific activities, ergonomic education.
De Buck et al. 2005	N=140; Rheumatic disease	Work status (job loss)	Usual care	No difference between groups in proportion of patients with job loss	I	Multidisciplinary	At least 2 visits in 4-12 weeks (total ± 3 h)	V	Multidisciplinary job-retention vocational rehabilitation using counselling and guidance, addressing identification of resources for adapting work environment, promotion of work self-efficacy.

Table 2 Characteristics of the included studies and interventions (*continued*)

Study characteristics		Intervention characteristics							
Detaillie et al. 2013	N=104; Chronic somatic disease	Work attitude	Usual care	No differences in self-efficacy and self-management after 8 months	G	Monodisciplinary	6 weeks with weekly 2.5-h sessions (total 15 h)	CB	Self-management program for workers with a chronic somatic disease, addressing skill mastery and goal setting, coping, positive reframing, communication, work situation.
Dorstyn et al. 2019	N=48; Spinal Cord Injury	Work attitude	Waiting list	No differences in job search self-efficacy after 4 weeks.	I	Multidisciplinary	4 weeks (online information, with weekly emails)	V	Online information package, involving six stand-alone learning modules which provided job-searching and career-planning information through text, videos, and interactive activities.
Feuerstein et al. 1993	N=34; MSD	Work status (RTW)	Usual care	Significantly more persons in IG than CG returned to work or were in vocational training after average of 17 months	GI	Multidisciplinary	4-6 weeks, daily 4-h sessions (total ± 120 h)	P	Multidisciplinary work re-entry rehabilitation program including warming-up, physical conditioning, work conditioning/simulation, job-related pain and stress management and ergonomic consultation.
Haffey et al. 1991	N=199; Traumatic brain injury	Work status (RTW)	Usual care	Differences were not tested. After 12 months 68% of IG and 34% of CG had paid employment	I	Multidisciplinary	3 months on average (total ± 60 h of staff assistance)	V	Work re-entry program, including vocational assessment, work hardening, job development, job analysis, job placement, transitional employment program, support and long-term follow-up.
Hammond et al., 2017	N=55; Rheumatoid Arthritis	Work status (job loss); Work productivity (sickness absence)	Written information	No significant differences between IG and CG after 9 months. The findings indicate VR may reduce risk of job loss and improve productivity.	I	Monodisciplinary	On average 4 hrs in 2 to 4 months	V	Vocational rehabilitation, starting with structured work interview and assessment of work barriers, followed by individualised programme including self-management at work, activity diaries, job accommodations. Also, written self-help information was provided.

Table 2 Characteristics of the included studies and interventions (*continued*)

Study characteristics		Intervention characteristics				
HUTTING et al. 2015	N=123; Chronic non-specific CANS	Work productivity; work attitude	Usual care	No differences between IG and CG	G	Monodisciplinary
Ipsen et al. 2012	N=297; Chronic condition	Work status (Employment)	(not described)	No differences in employment rate between groups	G	Monodisciplinary
Kendall & Thompson 1998	N=183; Chronic pain	Work status	Waiting list	Differences were not tested. After 15 months, 28.4% of IG and 9.8% of CG returned to work	G	Multidisciplinary
Keyser et al., 2018	N=287; MSD and rheumatic conditions	Work status (job loss); Work productivity	Written information	Significant fewer persons with permanent job loss in IG compared to CG after 2 years (p=0.03). No significant difference in WLQ scores between IG and CG after 12 months	I	Monodisciplinary
Li et al. 2006	N=64; MSD	Work attitude (work readiness)	Waiting list	Significant improvement in work readiness in IG compared with CG after 3 weeks	GI	Multidisciplinary

Table 2 Characteristics of the included studies and interventions (continued)

Study characteristics			Intervention characteristics						
LI-TSANG et al. 2008	N=63; MSD	Work status (RTW); Work productivity; Work attitude	Usual care	Significant higher RTW rate in IG than CG and significant differences in work readiness after 3 weeks. No differences in hours worked (WP) between groups	GI	Monodisciplinary	3 weeks with 4 1-h individual sessions +5 group sessions (total ± 14 h)	V	Job Placement and Support Program, including role play, case-management, structured job searching and preparation.
Macedo et al. 2009	N=32; Rheumatoid Arthritis	Work productivity	Usual care	No differences for work days missed. RA WIS significantly improved in IG compared to CG at 6 months	I	Monodisciplinary	6 months, with 6-8 sessions of 0.5-2h (total 3-16 h)	V	Comprehensive occupational therapy, including assessments (work, functional and psychosocial), ergonomic review; stress management; assertiveness, self-advocacy, activities of daily living, work visits.
MARHOLD et al. 2001	N=72; Chronic pain	Work productivity (No of days on sick leave)	Usual care	No differences between groups, after 6 months	GI	Monodisciplinary	12 weeks, with weekly 2.5-h sessions and 2 booster sessions (total 35 h)	CB	Cognitive behavioral return-to-work program addressing pain coping, goal setting, graded activity, pacing of activities, relaxation, cognitive techniques, stress management, problem solving.
Marnett & Selander 2000	N=47; MSD	Work status (level of benefit)	Usual care	Significantly more persons in IG than CG had lower level of benefits	GI	Multidisciplinary	8-17 weeks; 4 weeks daily 6-h sessions, followed by 4-13 weeks individual work training (total >120 h)	V	Extended multidisciplinary vocational rehabilitation program, including work training with case management addressing identifying problems, strengthen resources and self-confidence.
Martin et al. 2012	N=174; HIV/AIDS	Work attitude	Single session	No differences between IG and CG	GI	Multidisciplinary	7 weeks; 13 2-h sessions + 3 1-h individual sessions (total 29 h)	V	Mixed (group-individual) modality intervention that incorporated elements of motivational interviewing, skills building, job-related skills.

Table 2 Characteristics of the included studies and interventions (*continued*)

Study characteristics			Intervention characteristics				
MEYER et al. 2005	N=33; Chronic pain	Work status	Treat- ment by a physi- cian	No differences between IG and CG after 32 weeks	GI Multidisciplinary	8 weeks, daily sessions 3.5 h (5/ weeks) (total 140 h)	V Work rehabilitation program using an operant behavioral therapy approach, in- cluding education in ergonomics, learning strategies to cope with pain and to increase self-efficacy, workplace visit.
Niemeijer et al. 2010	N=71; Acquired brain injury	Work sta- tus; Work attitude	Waiting list	No differences in employ- ment rate between IG and CG after 6 months or in employability	G Monodisci- plinary	10 weeks, 20 sessions (total ± 20 h)	CB Vocational Transitions Program, address- ing overcoming obstacles, goal-setting, strategies for improving memory for work and daily living, self-awareness, stress management, problem-solving.
NTSIEA et al. 2014	N=80; Stroke	Work status (RTW)	Usual care	Significant higher RTW rate in IG than in CG after 6 months	I Multidisciplinary	6 weeks, weekly 1-h session and 1 4-h work skill assessment (total: 9 h)	V Workplace intervention program tailored to functional ability and workplace chal- lenges, performed at the patients' place of work and including interview with patient and employer separately to establish per- ceived barriers and enablers of RTW.
Ottomanelli et al. 2012	N=201; Spinal Cord Injury (SCI)	Work sta- tus; Work productiv- ity	Usual care	Significantly higher em- ployment rate in IG than in CG; persons in IG worked significantly more h/week	I Monodisci- plinary	SE services were provided during 1 year (mean 36 h)	V SCI Vocational Integration, based on Sup- ported Employment, including integrated vocational rehabilitation, job finding, com- petitive employment, ongoing job support, focus on participant preferences.
Salazar et al. 2000	N=120; Traumatic brain injury (TBI)	Work status (RTW)	Home rehab.	No differences between IG and CG in return to employ- ment	GI Multidisciplinary	8 weeks, daily program (group and individual therapies) (total ± 160 h)	V Intensive, standardized, 8-week in- hospital cognitive rehabilitation program. Group therapies addressed planning and organization, cognitive skills, pragmatic speech, psychotherapy and community reentry. Placement in various (military) work settings.

Table 2 Characteristics of the included studies and interventions (*continued*)

Study characteristics			Intervention characteristics			
Skouen et al. 2006	N=215; Chronic pain	Work status (RTW)	Usual care	Differences between IG and CG for total group were not tested; significantly fewer sick days for women in extensive program	GI Multidisciplinary P 4 weeks, 6-h daily program (5 days/week) (total 120 h)	Extensive multidisciplinary treatment, addressing cognitive coping strategies, body awareness training, occasional workplace intervention, lifestyle.
SULLIVAN et al. 2006	N=130; Chronic pain	Work status (RTW)	Physical therapy	Significantly more participants in IG than CG had returned to work after 4 weeks	I Multidisciplinary P Max. 10 weeks, weekly 1-h sessions (total 10 h)	Progressive Goal Attainment Program (PGAP) using cognitive techniques and physical therapy, maximizing activity involvement through activity monitoring, activity prescription, and graded activity participation.
TREXLER et al. 2016	N=44; Acquired brain injury	Work status (RTW)	Usual care	Significant more persons in IG than CG returned to competitive work, school or volunteering after 15 months	I Multidisciplinary V 15 months (individual needs, with mean 10.6 hrs, median 8.0 hrs)	Resource facilitation services, addressing patient and family education on brain injury, facilitation of community resources, vocational case coordination; also including outpatient rehabilitation therapies and neuropsychological services.
Varekamp et al. 2011	N=122; Chronic physical disease	Work status; Work attitude (self-efficacy)	Usual care	No differences in job maintenance or self-efficacy at work between IG than in CG	GI Monodisciplinary CB 20 weeks, 7 3-h group sessions + 3 individual sessions (total 24 h)	Job maintenance training program focusing on work related problems from an empowerment perspective, aiming to enhance knowledge, self-awareness and skills in order to help participants solve problems at work.
Van Vilsteren et al. 2017	N=150; Rheumatoid arthritis	Work productivity	Usual care	No effect on work instability or at-work productivity loss was found between IG and CG after 6 months	I Multidisciplinary V 12 weeks (individual needs; estimated 4-6 hr in total)	Care for Work program: Integrated care and participatory workplace intervention, based on participatory ergonomics.

Abbreviations: CANS= Complaints of arm, neck and/or shoulder, HIV=Human immunodeficiency virus, MSD=Musculoskeletal disorders; RTW= Return to work; Rehab=Rehabilitation; I=Individual therapy, G=Group therapy, GI=combined group and individual therapy; P=Physical focus; CB=Cognitive-Behavioral focus; V=Vocational focus; IG=Intervention group; CG=Control group; h=hour. (Author's name in capitals: study rated as high quality).

Effectiveness of interventions on work participation

A summary of the effectiveness and characteristics of interventions, related to the three outcome measures, is presented in Table 3. Twenty-two studies addressed one of the three outcome measures of work participation, seven studies addressed two, and one study addressed all three outcomes. In total, the 30 studies addressed 39 outcomes.

Below we describe the effectiveness for each of the three outcomes; these results are visualized in Figures 2-4.

Table 3 Characteristics and effectiveness of the interventions

		Number of studies (n=30)	Outcome ^a			Number of studies with significant effect (HQ) ^b (n=12 (5))	Number of studies with lack of effect (HQ) ^b (n=18 (3))
			Work status (n=22)	Work productivity (n=9)	Work attitude (n=8)		
Form							
Group + individual		11	8	3	4	5 (2)	6 (2)
Individual		13	10	4	1	6 (3)	7 (-)
Group		6	4	2	3	1 (-)	5 (1)
Approach							
Multidisciplinary		15	11	2	3	6 (4)	9 (1)
Monodisciplinary		15	11	7	5	6 (1)	9 (2)
Duration & intensity							
Short	High	5	5	-	-	1 (-)	4 (1)
	Low	9	4	2	6	4 (3)	5 (1)
Moderate	High	2	2	1	-	1 (-)	1 (-)
	Low	6	5	2	1	2 (1)	4 (-)
Long	Low	8	6	4	1	4 (1)	4 (1)
Focus of program							
Vocational		16	12	6	3	6 (3)	10 (1)
Cognitive-behavioral		9	5	3	5	3 (1)	6 (2)
Physical		5	5	-	-	3 (1)	2 (-)

^a: Total number of studies based on effectiveness on one of three outcomes: n=39 (work status: n=22, work productivity: n=9, work attitude: n=8)

Effects on work status (n=22)

Twenty-two studies measured the dichotomous difference in work status, such as return to work; (competitive, paid) employment, job maintenance, as outcome of the vocational rehabilitation intervention compared to the control condition (Table 3). Ten studies (45%) showed a significant difference in favour of the intervention and another seven (32%) a positive trend. The methodological quality of five studies (23%) was high, the quality of the other seventeen studies was judged as moderate or low.

The result of the meta-analysis of all studies including evaluation of the quality of the evidence are summarized in table 4 and presented in figure 2. When considering all twenty-two studies in the meta-analysis, we found evidence for a moderate effect of vocational rehabilitation interventions on work participation (RR 1.33; 95% CI 1.16-1.53). Rating down for indirectness was necessary, because there was considerable variation with regard to the content of the vocational intervention and the targeted patient populations. Although the lower border of the confidence interval was lower than 1.25 we did not rate down for imprecision, because the number of participants was very high. This means that we are moderately confident about this effect.

The result of the meta-analysis of five studies with high quality and low risk of bias is presented in figure 2. It shows a large positive effect of vocational rehabilitation interventions on work participation (350 participants; RR 1.57; 95% CI 1.26-1.97). Only rating down of one level for indirectness was necessary.

Effects on work productivity (n=9)

Nine studies (three of high quality), measured changes in work productivity (Figure 3, Table 3). Of these nine studies, six (67%) showed a positive trend in favor of the intervention, but the differences were small and not significant. The meta-analyses of all nine studies as well as the three high-quality studies yielded no significant differences (SMD 0.16 (95% CI -0.06-0.38) respectively 0.20 (95% CI -0.07-0.47). Rating down with three levels was necessary (risk of bias/inconsistency/indirectness, see Table 4), so the certainty of these results is very low.

Table 4 Evidence profile for the outcomes work status, work productivity, work attitude

Outcome (no. of studies)	Quality assessment					Number of patients		Quality of the evidence
	Limitation in design	Inconsistency	Indirectness	Imprecision	Publication bias	I ^a	C ^b	
Work status (22)	No serious limitation	No serious inconsistency	Indirectness	No serious imprecision	Unlikely	1221	1226	Moderate
Work productivity (9)	Limitation in design	Inconsistency	Indirectness	No serious imprecision	Unlikely	430	404	Very low
Work attitude (8)	Limitation in design	No serious inconsistency	Indirectness	No serious imprecision	Unlikely	278	240	Low

^a Number of patients who received the intervention studied.

^b Number of patients allocated to the control condition.

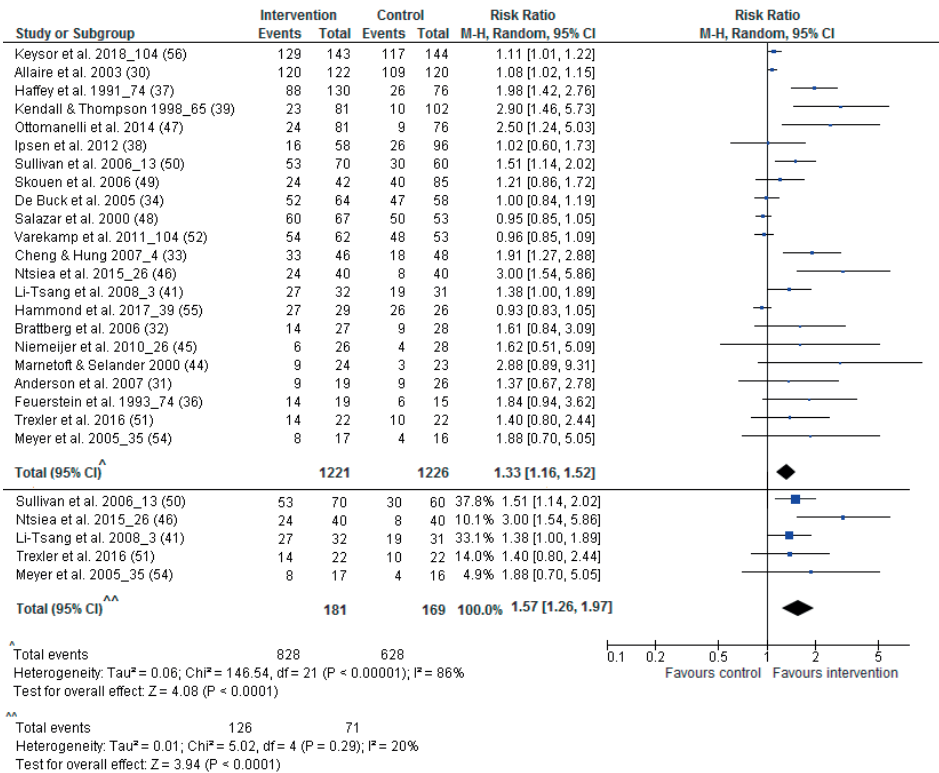


Figure 2 Effectiveness of interventions on outcome: Work status in all studies and High-Quality studies (ordered by sample size)

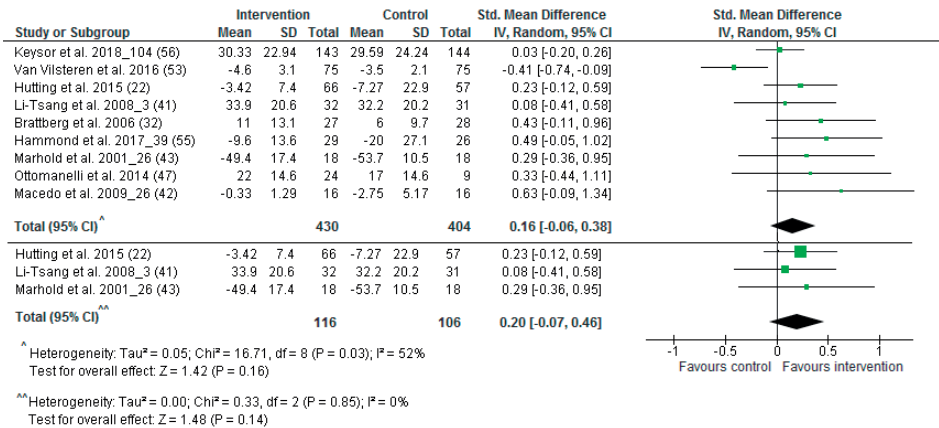


Figure 3 Effectiveness of interventions on outcome: Work productivity in all studies and High-Quality-studies (ordered by sample size)

Effects on work attitude (n=8)

Eight studies (three high quality) measured changes in work attitude (Figure 4, Table 3), of which two (25%) showed a significant effect in favour of the intervention and another three (38%) a positive trend. One study was not included in the meta-analysis because SDs were lacking [23]. When considering seven studies in the meta-analysis, we have evidence of an effect of small size (SMD 0.38; 95% CI 0.16-0.61) in favour of the intervention (n=518). The result of the meta-analysis including evaluation of the quality of the evidence is summarized in table 4. Rating down of two levels for risk of bias and indirectness was necessary, so we also have low certainty about this result. The result of the meta-analysis of three studies with high quality and low risk of bias is included in figure 4 and shows an effect of moderate size (SMD 0.59; 95% CI 0.18-1.00) in favour of the intervention (n=250).

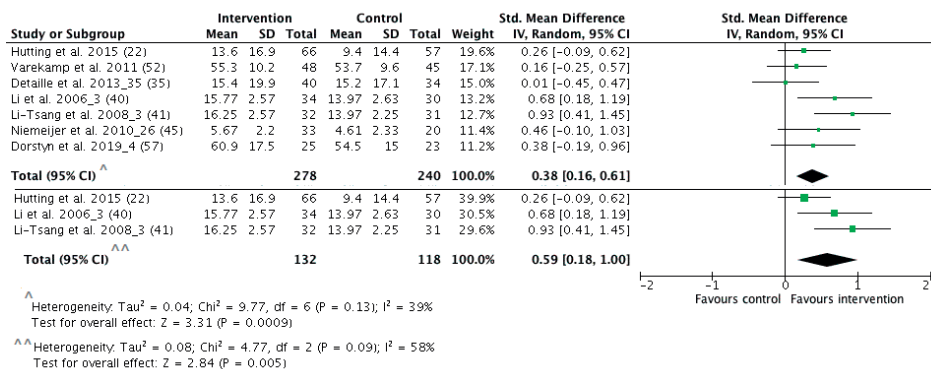


Figure 4 Effectiveness of interventions on outcome: Work attitude in all studies and High-Quality studies (ordered by sample size)

DISCUSSION

This systematic review summarizes data from 30 original studies in order to evaluate the characteristics and effectiveness of vocational rehabilitation interventions, that aim to improve work participation of persons with a chronic physical condition, other than back disorders. In contrast to the existing literature, we focused on interventions that apply a generic approach to enhance work participation of persons with a chronic physical condition, irrespective of the specific diagnosis. All interventions consisted of multiple treatment components, but varied widely regarding delivery characteristics and content.

Overall, 63-77% of the studies showed a positive trend on work participation, but only 25-45% of the studies showed a significant effect on work participation. Effectiveness of interventions was explored for three outcome measures of work participation: work status, work productivity and work attitude. After analyzing five trials with low risk of bias, we conclude with moderate certainty that there is a large positive effect of vocational rehabilitation interventions on work status. Analysis of both low and high quality trials ($n=22$) yielded a moderate positive effect with low certainty (RR 1.33). We conclude with low to very low certainty that vocational rehabilitation interventions also have a moderate positive effect on work attitude (SMD=0.59 or 0.39 respectively). We found no positive effects of vocational rehabilitation interventions on work productivity, although the certainty of this conclusion is very low.

All interventions in this review used a multicomponent approach; this makes it difficult to determine which component is (most) effective in enhancing work participation. Interventions that include multiple components are likely to be more effective in improving work participation than interventions consisting of a single component [10]. A multicomponent approach might be preferred because work participation is a complex process, requiring a balance between personal abilities, limitations and work demands. In persons with a chronic physical condition, the complexity of the process is increased by the unpredictable course and day-to-day variability of symptoms and, as a result, fluctuating personal physical abilities, that may interfere with the current work demands [8, 10, 58]. In addition, persons with a chronic condition have to balance work with other daily activities and life at home to be able to maintain work participation [8].

The results of this study, indicating a positive effect of the use of targeted interventions on enhancing work participation of persons with a chronic condition, are in line with systematic reviews addressing work outcomes for people with chronic back pain and mental health problems [11-13, 59, 60]. For people with chronic back pain, systematic reviews found evidence for a relevant effect of multidisciplinary interventions on return to work [11, 13] and a small effect of intense physical conditioning on reducing sick leave [12]. For persons with depression, adding a work-directed intervention to a clinical intervention reduced the number of days on sick leave; for individuals with severe mental illness, (augmented) supported employment was effective in obtaining and maintaining employment [59, 60].

Methodological considerations

This study provides a comprehensive and broad overview of vocational rehabilitation interventions across a wide range of chronic physical conditions, in contrast to most previous reviews that focused on a specific diagnosis. This broad overview is a potential

strength, because it increases insight in the generic components of vocational rehabilitation interventions and their effects on work-related outcomes.

We performed a systematic and comprehensive search in five databases containing the most important literature from biomedical and behavioural sciences, allied health and public health. However, due to the selection of certain databases, the exclusion of uncontrolled studies, and a restriction to the English language, we may not have identified all relevant trials. In line with our study objective, we used search terms addressing chronic physical conditions, as well as physical disability. Due to this generic approach, the literature search may not have included all studies addressing vocational rehabilitation in a specific diagnosis.

The analysis and synthesis of available evidence from the 30 studies was challenging due to the diversity of interventions, the different follow-up periods, and the lack of standardized outcome measures. In addition, the small sample sizes in a number of the included studies might have caused lack of power to detect intervention effects. Comparison of outcomes at different moments in time (ranging from 3-104 weeks) could also have influenced our results, because the achievement of positive effects on work-related outcomes requires a relatively long follow-up period. To improve transparency, we compared outcomes at 12-months wherever possible, and explicitly reported other follow-up periods in our results. Although included studies were heterogeneous, with respect to interventions, populations, and number of participants, we found it legitimate to rate down the level of evidence for the outcome work status with one level because the analysis of high-quality studies showed a heterogeneity of only 20%.

Finally, we included studies from 10 different countries. Since differing systems of disability benefits have an impact on rates of absence and return to work, these systems might have affected outcomes from the various countries in different ways [61].

Although only studies with a controlled study design were included, quality assessment of the included studies showed that about 30% of the studies had a low risk of bias, whereas the other studies had a higher risk of bias. The higher risk of bias of the majority of the studies was mainly due to selection bias (participants were less likely to be representative (referred from a source or self-referred), or low percentage of agreement to participate), blinding (assessors not being blinded or study participants were aware of the research question) and data collection tools not being valid and/or reliable. Evidence of low to very low certainty is a consistent finding in studies in the domain of vocational rehabilitation. Therefore, we performed meta-analyses of only high quality

studies as well as meta-analyses of both high and lower quality studies. When including only high quality trials in a meta-analysis, the effects were more pronounced.

Implications for practice

The overview of interventions in this review enables rehabilitation professionals to address work participation within rehabilitation using targeted interventions and making informed decisions about interventions. However, as a consequence of the perspective on a generic approach to improve work participation of persons with a chronic condition, and the broad variety of interventions and patient samples in the literature, the findings of this systematic review do not provide evidence which specific intervention will most effectively and efficiently benefit particular patients.

The main implications for clinical practice are twofold. First, addressing work-related problems within the rehabilitation care of persons with a chronic physical condition by means of a systematic and targeted intervention is beneficial to improve or sustain their work participation, with positive effects on finding or maintaining a job (work status) or return to work. Second, these beneficial effects seem to be generic, irrespective of the specific intervention characteristics or medical diagnosis. Notably, these generic effects were found for broad, multicomponent interventions that include individual support as one of the intervention components, whether or not combined with group sessions, and not for interventions with a single component only. Possibly, the individual support of patients may accommodate the fit between specific patient characteristics and work environment. Thus, selection of interventions should not be based on diagnoses, but professionals and persons with a chronic physical condition should select interventions that target the experienced barriers for work participation, such as physical limitations, coping with a chronic condition, or problems in the social work environment.

Implications for research

This systematic review reveals a need for further research on interventions targeting work participation in persons with a chronic physical condition to strengthen the evidence for effective interventions.

Future research should apply controlled study designs with high-quality methodology in order to improve the quality of evidence. Studies should apply a follow-up period of at least 12 months and preferably two years to examine outcomes on work participation. Primary outcome measures should be standardised, assessed and reported on all follow-up measurements. Furthermore, interventions should be adequately described to enable comparability of intervention components across studies and applicability

in practice. For this, the 12-item template for intervention description and replication (TIDieR) checklist can be used [62].

Finally, future research might provide evidence or increase insight in which specific intervention will most effectively and efficiently benefit particular patients to improve work participation.

CONCLUSION

Vocational rehabilitation interventions consist of multiple components, and vary widely regarding content and delivery characteristics. With information from 30 controlled studies, we conclude with moderate certainty that there is a large to moderate positive effect of vocational rehabilitation interventions on work status, and with low certainty that there is a moderate to small positive effect on work attitude. We found no effect on work productivity. Most studies showed positive effects or positive trends regarding work participation, irrespective of the specific intervention characteristics and diagnosis. The risk of bias in the majority of studies was high, implying that future studies with a lower risk of bias can add to the evidence for effectiveness.

COMPLIANCE WITH ETHICAL STANDARDS

This study complied with The Netherlands Code of Conduct for Scientific Practice from the Association of Universities in the Netherlands (VSNU). The requirements for authorship have been met and each author believes that the manuscript represents honest words.

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6

A vocational rehabilitation intervention for young adults with physical disabilities: participants' perception of beneficial attributes.

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ABSTRACT

Background: Finding and maintaining employment is a major challenge for young adults with physical disabilities and their work participation rate is lower than that of healthy peers. This paper is about a program that supports work participation amongst young adults with chronic physical disabilities. The study aims to explore their experienced barriers and facilitators for finding and maintaining employment after starting this program, the participant-perceived beneficial attributes of the program, and participants' recommendations for additional intervention components.

Methods: Semi-structured interviews ($n=19$) were held with former intervention participations. Interviews were recorded and transcribed ad verbatim. Themes were derived using the phenomenological approach.

Results: Physical functions and capacities, supervisor's attitude, self-esteem and self-efficacy, and openness and assertiveness were experienced barriers and facilitators for finding and maintaining employment. Improvement of self-promoting skills and disclosure skills through job interview-training, increased self-esteem or self-efficacy through peer-support, a suitable job through job placement, improvement of work ability through arrangement of adjusted work conditions, and change of supervisor's attitude through education provided to the supervisor were perceived as beneficial attributes of the intervention. Respondents recommended to incorporate assertiveness and openness skills training into future intervention programs.

Conclusions: The findings suggest that programs supporting work participation should be designed to provide challenging, real-world experiential opportunities that provide young adults with physical disabilities with new insights, self-efficacy and life skills. Also, such programs should facilitate context centered learning. Former intervention participants, therefore, evaluated job-interview training, sharing learning and social experiences with peers, job placement, arrangement of adjusted work conditions, and education as beneficial attributes of the 'At Work' program. In addition, they recommended, to incorporate more training on assertiveness and disclosure. We advise professionals to include these beneficial attributes in similar interventions in other contexts.

INTRODUCTION

Advancements in medical care result in increasing numbers of young people with chronic physical disabilities surviving into adulthood [1-2]. They have to develop the life skills and autonomy needed to participate in society [3-4]. However, studies showed that young people with physical disabilities report restricted autonomy and social participation [4-7]. The need for support in becoming independent adults is widely acknowledged [5, 8-9].

Finding and maintaining employment is a major challenge for young adults with physical disabilities [10-12] and their work participation rate is lower compared with healthy peers [12-14]. They experience barriers such as limited accessibility of buildings, discrimination, inadequate transportation, and lack of social support [10,15-17]. Improving their employment is important, because it provides financial independence, psychological well-being, social interaction, helps to develop self-identity and positively influences quality of life [18-19]. Healthcare teams and re-integration agencies have taken steps to support work participation amongst young people with physical disabilities. However, most of these interventions mainly address the development of life skills in general or provide support in finding temporary jobs during education [20-21]. To our knowledge, none of these programs primarily focus on support in finding and maintaining *regular* employment after post-secondary education.

To this aim, a multidisciplinary vocational intervention ('At Work') for young adults with physical disabilities is introduced in the Netherlands [22-23]. The intervention is designed for young adults entering the labour market, whether not employed or not suitably employed. Results of an evaluation of the first three groups ($n=12$) participating in this vocational intervention in the period 2007-2008 show that post-intervention and at three-years follow-up, two thirds of the participants were employed, with a paid/unpaid ratio of 1/1 post intervention and 7/1 at three-year follow-up [22]. In the Netherlands, a paid employment rate of 25% is an indicator for successful job coaching in this population, and thus the intervention was deemed feasible [22]. To date the 'At Work' program was only evaluated in a quantitative study, providing information on the effectiveness of this intervention. However, information on participants' experiences and beneficial attributes of the intervention is still missing. It is important to know the interventions elements that may work or may not work within programs. These insights will help to improve the 'At Work' program and will further enhance our understanding of beneficial attributes of similar interventions that can be applied in other contexts. This knowledge will improve the design and delivery of interventions that aim to foster work participation amongst young adults with chronic physical conditions after finish-

ing post-secondary education. Thus, this study aims to investigate the participants' perceived the following: i) barriers and facilitators for finding and maintaining employment after starting the 'At Work' program; ii) beneficial attributes of the program; and iii) recommendations for additional components within future intervention programs.

METHODS

Study design

A qualitative study was conducted using a phenomenological approach, to investigate the perceived beneficial attributes of the 'At Work' program [24]. Semi-structured interviews are particularly well suited to retrospectively explore former participants' experiences, in-depth thoughts and perspectives on barriers and facilitators for employment, beneficial attributes of the intervention, and recommendations for additional program components [25]. The Medical Ethical Committee of Erasmus University Medical Center approved the study (MEC-2012-381).

Intervention 'At Work'

A detailed overview of the content of the intervention is presented in Table 1. 'At Work' is a multidisciplinary vocational rehabilitation intervention for young people (16-27 years) with chronic physical disabilities [22-23]. This intervention provides support at finding and maintaining regular employment (not temporary or seasonal) after finishing post-secondary education. The 1-year intervention converges rehabilitation services provided by an outpatient rehabilitation clinic, and vocational services provided by a Re-integration Company. The Re-integration Company delivers re-integration and job-coaching services by order of a government-related insurance agency [22]. The intervention is especially designed for young adults with chronic physical disabilities, who (i) are not employed or not suitably employed, i.e. their employment do not fit with their educational level or interests, or is temporary, and they are looking for (more) suitable employment, (ii) finished post-secondary education or finish post-secondary education within 6 months, (iii) have an adequate understanding of the Dutch language, and (iv) have no severe cognitive or mental impairments.

The intervention starts with six two-hour group sessions, guided by an occupational therapist and a job-coach. A psychologist is involved in group sessions addressing psychological topics. The group sessions are primary aimed to increase participants' self-efficacy, knowledge, work-skills and awareness of their own values and needs. Group sessions entail peer-support, modelling, job interview skills training and education. Topics that are discussed during the group meetings consider work and health,

Table 1. Content of the 'At Work' program

	Aim	Content	Format	Trainers
Group-sessions				
Session 1	• Acquittance • To get insight into own attitude to work, and their own values and needs • To increase self-efficacy for work	Introduction	• Brainstorm • Role-model • Peer-support	• Occupational therapist • Jobcoach
Session 2	• To get insights into interests and capacities for work • To set goals on work participation, self-care and leisure activities	Work motivation: interests and capacities	• Group-discussion • Peer-support • Goal-setting	• Occupational therapist • Jobcoach
Session 3	• To get insight into own coping style • To be aware of different ways of dealing with a chronic condition at work • To increase self-efficacy for work	Work and health	• Assessment of coping style • Group-discussion • Peer-support • Role-model	• Occupational therapist • Jobcoach • Psychologist •
Session 4	• To educate young adults about social benefits and insurances • To be able to get the needed information • To increase self-efficacy for work	Social benefits and insurances	• Education	• Occupational therapist • Jobcoach
Session 5	• To be able to search for vacancies • To be able to write an application letter and Curriculum Vitae • To increase self-efficacy for work	Searching for vacancies	• Peer-education • Peer-support • Group-discussion • Education	• Occupational therapist • Jobcoach
Session 6	• To train job-interview skills • To increase self-efficacy for doing a job-interview	Job interview training	• Job-interview skills training • Peer-support	• Occupational therapist • Jobcoach
Individual coaching				
	• To find suitable employment • To sustain employment	• Job placement • Arrangement of adjusted work conditions • Education provided to supervisor	• Work skills training • Education	• Jobcoach • Occupational therapist (If nesserary)

interests and capacities, social benefits and insurances, searching for vacancies and doing a job interview. After group sessions, the intervention is continued with weekly individual coaching sessions provided by a job-coach for about 1 year. Individual coaching entails help in finding a job, and support at dealing with the social and physical work environment, i.e. informing the participant's employer and colleagues about the chronic condition and arrangement of adjusted work conditions.

Study population

Of the 59 persons enrolled in the intervention program in the period 2007-2011, 37(63%) completed the group-sessions and individual coaching. Eleven participants dropped out during group sessions because of the progressive severity of their physical condition ($n=2$), psychosocial problems ($n=1$), time constraints ($n=2$), or unknown reasons ($n=6$). Another 11 participants did not start or maintain the individual coaching because of progressive severity of their physical condition ($n=1$), no finished post-secondary education ($n=1$), low self-esteem ($n=1$), no motivation for work ($n=1$), negative experiences during job placement ($n=1$), other treatment goals ($n=1$), no financial benefits for job coaching ($n=2$), or unknown reasons ($n=3$).

For the semi-structured interviews we selected respondents using a total sample approach [26]. That is, we sent an information letter to all the 37 participants who had completed the full program. The researcher Marjolijn Bal (MB) answered any questions of those respondents who expressed interest in participating on a reply form by telephone, before asking them to sign the informed consent form.

Data collection

Background characteristics

From the start of the intervention the Re-integration Company systematically monitored data for the pilot study in 2007-2009 [22], and thereafter continued systematic data-monitoring for research purposes. Two researchers Marjolijn Bal and Natascha van Schaardenburgh (MB&NS) retrospectively reviewed the charts of all included participants ($n=37$). Data were systematically extracted from the clients' charts and entered in a digital data extraction form [27]. Chart review took place from March 2013 till February 2014.

The following background characteristics were reviewed from the charts: age, gender, medical diagnosis, educational level (three levels, referring to actual and potential educational capacity) [13] and type of education (special or mainstream). The severity of the physical limitations was assessed using the physical limitations indicator developed by the Organisation for Economic Cooperation and Development (www.oecd.org). The scale

assessed physical limitations and limitations in hearing and seeing, and is considered as reliable and valid [28]. We collected data on work status (paid and unpaid employment, unemployment) at pre and post-intervention. Employment was defined as performing a job for ≥ 12 h/week, in accordance with the definition of Statistics Netherlands [29].

Interviews

The interviews consisted of three steps, using an interview guide. First, interviewees were asked to indicate their perceived barriers and facilitators for finding and maintaining employment after starting the intervention. Second, the respondents were asked to indicate the beneficial attributes of the 'At Work' program. Last, they were asked to indicate, if relevant, some recommendations for additional program components. Perceived barriers and facilitators for employment were used as starting point for their reflection on beneficial attributes of the 'At Work' program, and the recommendations for additional program components. During the interviews, the trained and experienced interviewer (MB) introduced the topics following the natural course of the conversation, and allowed open-ended conversation to continue until, in her judgment, each topic was exhausted. The interviewer worked as a researcher and teacher in higher education. Interviews were conducted at an outpatient rehabilitation center at a time convenient to the participant. Interviews lasted between 50 and 90 mins, with an average of 60 min each. After each interview field notes were made. Interviewing and data analysis were alternated; interviews were analyzed after each set of three interviews. Respondents were well informed about the research goals. However, there was no relationship between researcher and respondents prior to study commencement. Interviews were held between November 2012 and January 2013.

Data analysis

Non-response analysis was performed using a Chi-square test for categorical variables and independent t-tests for continuous variables in order to detect differences in age, gender, onset of diagnosis severity of limitations, level of education, type of education, and employment status between interviewees and non-interviewees. McNemar tests were used to compare employment rates pre and post-intervention within the sub-groups total sample, interviewees and non-interviewees.

Interviews were audio taped, transcribed verbatim and imported into Atlas.ti (6.2). The analysis started with entirely reading all transcripts, in order to explore the data. Based on the interview guide data were categorized into three main parts: perceived barriers and facilitators for finding and maintaining employment after starting the intervention, perceived beneficial attributes of the 'At Work' program, and recommendations for additional program components. Thereafter, two researchers Marjolijn Bal and Jane Sat-

toe (MB&JS) with different professional backgrounds, i.e. respectively psychology and health sciences, independently applied an inductive process of thematic analysis within the three main parts of the interviews in order to detect codes. Themes were, hereby, empirically derived from the data [24, 30]. After analyzing a set of three interviews the researchers discussed their individual codes in order to reach consensus about the final codes. Through this process of organizing and comparing codes, key themes were identified [24]. The researchers' triangulation enhanced the trustworthiness of the findings [31]. Comparing and discussing the independently applied inductive analysis enhanced confirmability of findings. Also, the different educational backgrounds of researches reduced discipline specific biases, which enhanced the transferability of the study results. Final codes were reviewed and discussed with all research team-members. The research team-members had first-hand experience with the program (NS&MF) and knowledge regarding, childhood disability, youth transitions and work participation (Marij Roebroek, MR, Harald Miedema, HM). Team-members therefore were familiar with the context of the intervention being explored, which contributed to the credibility of the themes derived [30]. To provide a systematic overview of the experienced barriers and facilitators for employment, two researchers (MB&MR) classified them according to the main components of the International Classification of Functioning, Disability and Health (ICF), i.e. Body functions and Structures, Activities and Participation, Environmental Factors and Personal Factors [32]. In a case of disagreement, the ICF component was discussed with the other research team-members to reach consensus.

RESULTS

Study sample

Background characteristics of the study sample are presented in Table 2. Interviewees and non-interviewees did not significantly differ in age, gender, type of chronic condition, onset of chronic condition, severity of physical limitations, educational level and pre-intervention work status. Significantly more interviewees (68.4%) have followed special secondary education youth with physical disabilities compared with non-interviewees (33.3%), ($\chi^2(2)=4.555, p<.05$). The paid employment rate of interviewees increased over time from 5.3% pre-intervention to 36.8% post-intervention ($p>.05$). All respondents that indicated their interest for participation into this research were interviewed ($n=19$). After fifteen interviews no new relevant codes emerged from data-analysis.

Table 2. Background characteristics of the study sample

	Intervention participants in the period 2007-2011 (N = 37) <i>Mean ± SD / Frequency</i> (%)	Subsample interviewees (N = 19) <i>Mean ± SD /</i> <i>Frequency (%)</i>	Subsample Non-interviewees (N = 18) <i>Mean ± SD /</i> <i>Frequency (%)</i>
Age	26.0 ± 2.7	25.8 ± 2.3	26.2 ± 3.1
Gender (<i>male</i>)	19 (51.4)	8 (42.1)	11 (61.1)
Diagnoses			
Nervous system			
Cerebral palsy	11 (29.7)	6 (31.6)	5 (27.8)
Acquired brain injury	7 (18.9)	4 (21.1)	3 (16.7)
Multiple sclerosis	3 (8.1)	1 (5.3)	2 (11.1)
Spina bifida/Spinal tumor	3 (8.1)	2 (4.9)	1 (5.6)
Epilepsy	2 (5.4)		2 (11.1)
Muscular diseases	2 (5.4)	1 (5.3)	1 (5.6)
Musculoskeletal disorders			
Rheumatod arthritis	2 (5.4)	2 (10.5)	
Scheuermann's disease	1 (2.7)		1 (5.6)
Osteogenesis imperfecta	1 (2.7)		1 (5.6)
Other			
Sickle-cell anemia	1 (2.7)		1 (5.6)
Hodgkin	1 (2.7)	1 (5.3)	
Chronic pain	1 (2.7)	1 (5.3)	
Asthma	1 (2.7)	1 (5.3)	
Diabetes mellitus	1 (2.7)		1 (5.9)
Onset chronic condition (<i>congenital</i>)	16 (43.2)	9 (47.4)	7 (38.9)
Severity physical limitation			
No	7 (18.9)	2 (10.5)	5 (27.8)
Slight	18 (48.6)	9 (47.4)	9 (50.0)
Moderate and severe	12 (32.4)	8 (42.1)	4 (22.2)
Educational level			
High	6 (16.2)	4 (21.1)	3 (16.7)
Middle	25 (67.6)	12 (63.2)	13 (72.2)
Low	6 (16.2)	3 (15.8)	2 (11.1)
Special education (<i>yes</i>)	19 (51.4)	13 (68.4)	6 (33.3)*
Work status			
Pre-intervention			
Total employed (Yes)	17 (45.9)	8 (42.1)	9 (50.0)
Paid (Yes)	7 (18.9)	1 (5.3)	6 (33.3)
Unpaid (Yes)	10 (27.0)	7 (36.8)	3 (16.7)
Not employed (Yes)	20 (54.1)	11 (57.9)	9 (50.0)
Post-intervention			
Total employed (Yes)	24 (70.6)	12 (63.2)	12 (80.0)
Paid (Yes)	15 (44.1)**	7 (36.8)	8 (53.3)
Unpaid (Yes)	9 (26.5)	5 (26.3)	4 (26.7)
Not employed (Yes)	10 (29.4)	7 (36.8)	3 (20.0)

* Differs significantly from the interviewees $p < .05$; ** Significantly increased over time, within the total group of intervention participants $p < .05$

Experienced barriers and facilitators for finding and maintaining employment

From data-analyses on this part of the interviews, the following themes emerged: physical functions and capacities, supervisor's attitude, the young adult's self-esteem and self-efficacy, and openness and assertiveness. Participants identified these themes after starting the intervention.

Respondents indicated that they could not apply to all vacancies they were interested in because of their **physical functions and capacities**. Also they mentioned that they were not able to fulfil all job tasks. A male respondent (age 26, cerebral palsy) for instance explained: *"My work ability is much lower than hundred percent. I was hired at a package service company. One of the tasks was to pack things. I worked very hard at that company. But after one year they (the employer) said you cannot do this task with your one hand. I said what did you expect? [...] I can put the bottle and soap together. However, do not ask me to pack it. I cannot do that with only one hand"*.

Importantly, according to the respondents, a **supervisor's positive attitude** increases chances to get and maintain a job. That is, being open minded and showing understanding for their specific situation. Respondents who sustained their employment mentioned that employers agreed with flexible working hours and tasks, allowed for doctor appointments, and made buildings and toilets accessible. As a female respondent (age 25, spina bifida) for example mentioned: *"In the beginning my supervisor assigned me job tasks that I could not fulfil because of my physical disabilities. At one moment he asked me: May I help you? He came up with a solution and now asks me more suitable tasks to do"*.

Many respondents perceived **self-esteem and self-efficacy** as facilitator for their search for employment and for sustaining employment. A female respondent (age 30, muscular disease) for instance explained: *"You have to emphasize your capacities and not be too modest."* Another female respondent stated: *"For some people it is hard to look the employer straight in the eyes. Well, I do. People with disabilities should believe that they have the necessary skills for social participation. I believe in my skills"* (age 25, acquired brain injury).

Respondents indicated not being **open** and **assertive** as barriers for maintaining employment. Not being open and assertive makes it difficult to talk to the employer, to ask for help, to inform colleagues about the condition, and to indicate specific needs, as the respondents noted. *"You feel happy to have a place to do your internship, so you do not want to tell them that you can only work for 20 hours. I worked for 40 hours and managed*

to keep this up for almost two weeks. [...] However, they now pay attention to that aspect and tell me to go home if my working time is over” (female respondent, age 25, acquired brain injury).

Participant-perceived beneficial attributes of the ‘At Work’ program

Themes emerged from data-analyses considering perceived beneficial attributes were as follows: job-interview training, job placement, arrangement of adjusted work conditions, peer-support, and education provided to the supervisor.

Respondents perceived the **job interview training** as beneficial attribute of the ‘At Work’ program, because the job-interview training helped them to develop self-promoting and disclosure skills. The self-promoting and disclosure skills, in turn, covered the barrier for employment physical functioning and capacities. A female respondent for instance indicated the following: *“Before I started the intervention I send twenty application letters, but I was rejected for all the jobs because of my limited physical capacities. During the intervention I learned how to promote myself. Now I know my strengths and difficulties and I learned to be focused on my strengths”* (age 24, rheumatoid arthritis). A female respondent (age 21, chronic pain) for instance, explained: *“I learned a lot from the job interview training. For instance, I learned how I could answer some questions about my chronic condition. Also, I learned how to say things differently”*.

Respondents indicated that the intervention addressed the barrier limited physical functioning and capacities. They mentioned the **placement on suitable jobs and arrangement of adjusted work conditions** as beneficial attributes of the intervention, because these were perceived to provide them suitable jobs and improve their work-ability. *“I am allowed to apply for all kind of jobs I like. However, jobs should be tailored to my physical capacities, and I do not know the jobs I could apply for. The job coach, conversely, has a great social network and came up with suitable vacancies. [...] Now, I am able to do my work very well, despite my disability”* (male respondent, age 27, spina bifida).

Respondents mentioned that the intervention covered the barrier or facilitator for employment supervisor’s attitude. They indicated the **education provided to the supervisor** about the nature of the chronic condition and the availability of social security benefits as beneficial attribute of the intervention, because it positively changed their supervisor’s attitude. For instance a female respondent with acquired brain injury (age 26) stated: *“When I was not able to explain something about my chronic condition, my job coach provided the necessary information about my chronic condition to my employer. Consequently the understanding increased and people were more willing to provide help”*.

Respondents acknowledged that the intervention positively influenced their self-esteem and self-efficacy. A respondent (female, age 25, rheumatoid arthritis) for instance explained: *“I did not feel alone any longer [...] and it helped me to be more confident about myself”*. For almost all young adults in the program, the improved self-esteem and increased self-efficacy came from other group members. Participants perceived **peer-support** as a beneficial attribute of the intervention, as the following female respondent (age 24, rheumatoid arthritis) for instance indicated: *During the group-sessions I met other people with disabilities. I realized, that I was not the only one with a disability. We shared experiences and gave each other advice. [...] My self-confidence increased”*

Participants’ recommendations for additional program components.

From data-analyses on these parts of the interviews, the following themes emerged: role models, and openness and assertiveness skills training.

Participants indicated the improvement of their self-esteem as benefit of the intervention. However, they did not perceive **the use of role models** as very helpful. A male respondent (age 25, acquired brain injury), for instance, explained the following: *“The role model did not help me to feel more confident about myself. I could not compare myself with him. [...] He [the role model] had a muscular disease since he was born. Consequently he could prepare for his transition to work his whole life. For me it is different. I had a normal life, which suddenly fell into pieces”*.

Some respondents stated that they did not learn during the intervention to be assertive and open at work, and the lack of these skills barriers their employment. Therefore they brought forward the **need for openness and assertiveness skills training** as additional intervention component. For example: *“A course or workshop that trains you how to deal with colleagues asking about your condition. It would allow you to be clearer in what you can do and what you cannot do. In what you like and in what you dislike. I think that is important, because your colleagues do not read your application form. So, they will ask, and you will have to explain it to them. That is where it all starts”* (female respondent, age 25, cerebral palsy).

Table 3 provides a systematic overview of the findings that emerged from qualitative data-analyses.

Table 3. Systematic overview of themes emerged from qualitative data-analyses

Experienced barriers or facilitators for finding and maintaining employment, classified according to the International Classification of Functioning, Disability and Health		Participant-perceived beneficial attributes of the intervention		Participant-provided recommendations for additional or adjustment of intervention components
ICF component	Experienced barrier or facilitator	Attribute	Benefit	
Activities and Participation	Physical functions & capacities	Job-interview training (group-sessions)	Development of a) self-promoting skills and, b) disclosure skills, i.e. skills needed to be open about the chronic condition	
		Job placement, i.e. support at finding jobs, that fits with their physical capacities (individual coaching)	Suitable job	
		Arrangement of adjusted work conditions (individual coaching)	Suitable job/ improved work ability	
Environmental factors	Supervisor's attitude	Education of supervisor about the chronic condition and social security benefits (individual coaching)	The supervisor's negative attitude changed into a positive attitude	
Personal factors	Self-esteem and self-efficacy	Peer-support (group-sessions)	Self-esteem and self-efficacy increased	Role-models
Personal factors	Openness and Assertiveness [^]	None	None	Openness and assertiveness skills training (group-sessions)

[^] Openness and assertiveness were only mentioned as barriers for *maintaining* employment.

DISCUSSION

The present study sheds light on beneficial attributes of an intervention that provides support on finding and sustaining employment after finishing post-secondary education amongst young adults with chronic physical conditions. Participant-perceived beneficial attributes of the program were job-interview training, job placement, arrangement of adjusted work conditions, education, and peer-support. These beneficial attributes were perceived to address experienced barriers and facilitators for finding and maintaining employment, and these barriers and facilitators were also found in earlier studies [10-11]. According to the former intervention participants, however, openness and assertiveness skills training should also be incorporated into the intervention.

The opportunity for sharing learning and social experiences with peers during the group session of the 'At Work' program was considered to lead to changes in self-efficacy. Results of our study underlined the importance of incorporating real-world experiential opportunities into interventions that aim to provide young adults with new insights, and foster self-efficacy and life skills. The importance of opportunities for real-world experiences and context centered learning is mentioned before with regard to life skills development and empowerment of adolescents with chronic conditions [33]. Our study showed that these program elements are also beneficial for young adults entering the labor market after finishing education. The relation between work participation and self-efficacy was mentioned in previous studies on adults with physical disabilities [34-35]. Although modelling is assumed to be an appropriate strategy to foster people's self-efficacy, respondents did not evaluated it as valuable [36-37]. Respondents could not identify themselves with the models. This underlined the importance to carefully select role models for the group sessions, perhaps even tailored to the types of physical disability amongst the participants [38]. Currently, former intervention participants are invited into the intervention as role models. Alternatively, participants' self-efficacy could be fostered by other strategies, for instance identifying their personal barriers and practicing new coping strategies [37].

Also, respondents indicated the importance of disclosure and assertiveness skills for finding and maintaining employment and they would have welcomed more training on these skills. Assertiveness and communication skills are examples of life skills [38] and a lack of critical life skills is recognized as factors that may contribute to difficulties young people with chronic physical conditions face in their social participation [4,39]. The importance of assertiveness and openness for finding and maintaining employment for adults and adolescents with chronic conditions and the need for support on these issues is also earlier mentioned in literature [35,40]. Our study showed that this also applies

to young adults with chronic physical conditions searching for regular employment. Kingsnorth et al., 2014 [41] provided an overview of content and effectiveness of general life skills programs. Most programs that are focused on improvement of assertiveness skills incorporated role playing as core element into the intervention. In line with the social learning theory [42], a role play between an employee (intervention participant) and employer (actor) could be a learning experience for the young adult that plays the role of employee as well as the other intervention participants. Role playing, thus, could be an appropriate format to actively train assertiveness skills and may be incorporated into the 'At Work' program.

Although the respondents mentioned the importance of life skills development, respondents highly valued the effort of the job-coach on the arrangement of specific work conditions, the adjustment of the physical work environment and on the supervisor's attitude. These experiences address an important issue described in the literature about social participation, namely the interactions between the person and environment [43-45]. Barnes & Mercer (2005) [46] argued that the exclusion of people with chronic conditions from employment opportunities can be largely attributed to the social work environment rather than to individual impairments. Thus, the question arises whether we should more intensively intervene on the social (work) environment. Employers or supervisors may also be invited to participate in additional and more comprehensive training programmes, as to equip young adults with the necessary skills and knowledge to facilitate sustainable employment [47].

Strengths and limitations of the study

To our knowledge this is one of the first studies that used participants' lived experiences to evaluate the perceived benefits of a vocational intervention, and the participants' recommendations for additional intervention components. Qualitative research methods may shed light on how participants' context interacts with experiences, and thus provide some insights about perceived benefits and needs of an intervention to improve work participation of young people with chronic physical conditions [37]. This helped to identify 'effective ingredients' of interventions and answer the question of what works to improve regular employment amongst young adults with physical disabilities [48].

The study included a heterogeneous sample of young adults with a wide range of rather severe chronic physical disabilities that were either congenital or acquired. It is not possible, therefore, to relate findings to specific diagnoses. However, because all young adults with chronic physical conditions entering the labour market may face similar adaptive challenges, studying a heterogeneous sample can be considered as a strength

[49]. This assumption is supported by the fact that participants recognized each other's experiences during group sessions.

The small study sample of the qualitative study could be another limitation, because it might limit the generalization of the study results. However, interviewees did differ only significantly from non-interviewees with respect to a higher percentage with a background in special education. Although this might have influenced the present study results [50], according to the study participants not the type of education influenced their present employment, but their level of education. The latter was similar between both subgroups.

Finally, at the time of this study 'At Work' was only delivered at one rehabilitation centre. At present, the intervention is implemented in other rehabilitation centres, and is currently evaluated using a multicenter controlled study design (Dutch Trial Register, NTR=4145).

CONCLUSION

There is a need to support young adults with physical disabilities to develop necessary life skills and to overcome barriers for finding and maintaining employment after finishing post-secondary education. The present findings suggest that vocational rehabilitation programs should be designed to provide challenging, real-world experiential opportunities that provide young adults with new insights, self-efficacy and life skills. Also, these programs should facilitate context centered learning. Former intervention participants, therefore, evaluated job-interview training, peer-support, job placement, arrangement of adjusted work conditions, and education as beneficial attributes of the 'At Work' program. In addition, they recommended, to incorporate more training on assertiveness and disclosure. We advise professionals to include these beneficial attributes in similar interventions in other contexts.

KEY MESSAGES

- Former intervention participants evaluated 'At Work' as a promising and valuable intervention to improve work participation among young adults with physical disabilities who finished education.

- These young adults underlined the importance of challenging, real-world experiential opportunities that provide them with new insights, self-efficacy and life skills, and providing context centered interventions for improving their work participation
- Former intervention participants, therefore, indicated the job-interview training, peer-support, job placement, arrangement of adjusted work conditions, and education as beneficial attributes of the intervention
- Participants recommended to incorporate more training on assertiveness skills and disclosure into this intervention or similar vocational programs, so as to equip them with the necessary social skills to deal with the social and physical work environment, and to sustain their regular employment after finishing post-secondary education.

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7

Entering the labor market: Increased employment rates of young adults with chronic physical conditions after a vocational rehabilitation program

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ABSTRACT

Purpose: Employment of young adults with chronic physical conditions entering the labor market after finishing post-secondary education remains behind compared to typically developing peers. The aim of this study is to evaluate changes in their paid employment levels after following a vocational rehabilitation intervention ('At Work').

Materials and methods: Participants aged between 16 and 27 years ($n=90$) were recruited via rehabilitation physicians and a jobcoach agency and participated in a vocational rehabilitation program. Cochran's Q and McNemar tests served to test the development of intervention participants' paid employment over time. Chi-square tests were used to compare intervention participants' paid employment level with national reference data selected on age and having a self-reported chronic physical condition.

Results: Paid employment level of the intervention cohort significantly increased from 10.0% at baseline to 42.4% at 2-years follow-up ($p < 0.001$). At 2-years follow-up, their employment rates approached the employment rates of national reference data (42.4% versus 52.9%, $p = 0.17$).

Conclusion: Starting from a disadvantaged position, the paid employment rate of the intervention cohort substantially increased over time, approaching the employment rate of reference data. 'At Work' seems to be appropriate for supporting this specific group who face obstacles to enter the labor market, to find competitive employment.

INTRODUCTION

Advancements in medical care enable the vast majority of children with chronic physical conditions to live into adulthood [1, 2]. Emerging adulthood represents a critical developmental stage for all young people as they experience multiple transitions including leaving high school, finishing post-secondary education, getting a job, forming new personal relationships and moving out of parents' homes to live on their own [3, 4]. Previous studies consistently report that young people with chronic physical conditions more often experience restricted autonomy and societal participation [3, 5, 6], and need support in developing adult roles [3, 7-9]. Roebroek and colleagues underlined the need for incorporating a lifespan perspective throughout pediatric and adult healthcare [8]. From this perspective, Young Adult Teams of health care professionals have been set up, which proved to be useful in supporting young adults' social participation, for instance by providing group-based interventions focusing on specific life areas [8, 10].

In the process of transition into adulthood, one of the challenges is to find work [11-14]. Young adults with chronic physical conditions experience substantial difficulties, such as problems with disability disclosure, traveling to work, limited accessibility of buildings, limited adaptation of workplaces or working conditions, discrimination, lack of support of manager or colleagues, low self-esteem and lack of life skills [9, 12, 15-19]. In persons with chronic physical conditions, low employment rates are common in both Europe and the USA [20, 21]. Adequate support in finding and keeping work may result in optimal work participation, and may prevent lifelong financial dependency, unemployment, and poor quality of life [22, 23].

Several interventions are developed to support young people with chronic physical conditions in finding and maintaining employment. Most of these interventions mainly address the development of more general life skills or provide support in finding temporary jobs as a student [24-29]. To our knowledge, none of these programs are primarily focused on support for entering the labor market after finishing post-secondary education, and to this aim the 'At Work' program was developed [24, 25]. 'At Work' is a vocational rehabilitation program provided by the Young Adult Teams of three out-patient rehabilitation clinics in The Netherlands. The intervention includes a combination of group education, peer-mentorship, individual jobcoaching based on the supported employment methodology, and experimental learning. These intervention elements are evaluated as useful and appropriate to improve psychosocial development and vocational participation among young people with chronic conditions [24, 25, 28-31]. Studies on existing programs that address the development of life skills in general or provide support in finding temporary jobs during education, showed some positive preliminary

results [25-29]. However, there are relatively few rigorously designed, published studies that have evaluated the effectiveness of life skills and vocational rehabilitation programs for young adults with physical disabilities, and there is a need for large-sample studies in this field [25, 28]. So far, also the 'At Work' program was only evaluated in a feasibility study ($n = 12$), indicating increased paid employment rates from 8% at baseline to 33% and 42% at 1-year and 2-years follow-up [32].

Therefore, the current study aims to evaluate changes in employment rates, in a large cohort of participants of the 'At Work' program. Employment rates at baseline (T0), post-intervention (T1) and 2-years follow-up (T2) were assessed for intervention participants and compared to external national reference data of young adults with chronic physical conditions. Due to the disadvantaged starting position of intervention participants - since they entered the intervention because of difficulties in finding and maintaining competitive employment - we assumed that, at the start of the intervention the paid employment rates within the intervention cohort were lower compared to those of the national reference data. We hypothesized that a) within the intervention cohort, the rates of paid employment will significantly increase in 2 years' time; and b) the paid employment rates of the intervention cohort at 2-years follow-up after baseline will equal the rates of the reference data of young adults with chronic physical conditions, thus overcoming their disadvantaged employment participation.

MATERIALS AND METHODS

Design

Cohort study with 2-years follow-up after baseline, and comparison with external national reference data selected on age (16 - 30 yrs) and having a self-reported chronic physical condition.

Vocational rehabilitation intervention: 'at work'

'At Work' is a multidisciplinary vocational rehabilitation intervention for young people (16-27 years) with chronic physical conditions. Participants have finished their post-secondary education and experienced difficulties in finding and/or maintaining competitive employment, i.e., not seasonal or temporary. The intervention aims to improve young adults' life skills and to provide support for their vocational participation [32, 33]. A detailed overview of the content of the intervention is presented elsewhere [30].

The 1-year program integrates vocational services provided by a job coach of a reintegration company into rehabilitation services provided by a Young Adult Team of an out-

patient rehabilitation clinic. The intervention consists of six two-hour group sessions, facilitated by an occupational therapist and a job coach. A psychologist is involved in some group sessions addressing psychological issues. Group sessions aim to empower young adults and, thus, increase participants' self-efficacy, knowledge, work-skills and awareness of their own values and needs [34, 35]. Participants learn to apply for a job, to negotiate for workplace modifications or adjustment of working conditions, to disclose their condition and to feel self-efficacious in dealing with work-related problems. Group sessions entail peer-support, modelling, experimental learning and education. These intervention elements are evaluated as useful and appropriate to improve young adults' life skills and to provide support for their vocational participation [24, 25, 28-30]. Topics that are discussed during the group meetings consider work and health, interests and capacities, social benefits and insurances, searching for vacancies and performing a job-interview. The intervention continues with weekly individual coaching sessions based on the supported employment methodology [31]. These sessions are provided by a job coach for about one year. This coaching entails help in finding and applying for a suitable job, and support for dealing with the social and physical work environment, i.e., informing the employer and colleagues about the chronic physical condition, and arrangement of workplace modifications or adjusted working conditions. If necessary, an occupational therapist could be consulted during the individual coaching trajectory.

Recruitment intervention cohort

The vocational rehabilitation intervention was developed in 2007, and from that point until now provided at outpatient rehabilitation clinics with Young Adult Teams. In the current study we included all young adults that participated in the intervention in the period 2007-2016. Three rehabilitation centers participated: Rijndam Rehabilitation/ Erasmus MC (Rotterdam), Reade Rehabilitation Centre (Amsterdam) and Basalt (Leiden). These centers collaborated with the job coach agency 'VolZin'. Young adults with chronic physical conditions were referred to the intervention by a rehabilitation physician, if they met each of the following inclusion criteria: a) aged between 16 and 27 years, b) unemployed or not suitably employed, defined as a job that is not consistent with the individual's education, physical abilities and preferences, c) had finished education, or will finish education within 6 months; d) an adequate understanding of the Dutch language and d) no severe intellectual impairments. All participants received verbal and written information about the study. If they were willing to participate in the study they signed an informed consent form. The medical ethics committee of Erasmus MC Rotterdam approved this study (MEC-2012-381) and all participating centers granted local approval.

Measurements intervention cohort

Procedure

Data of intervention participants in the period 2007-2009 were collected as part of a prospective feasibility study [32, 33] using self-report questionnaires. Data of participants in the period 2009-2012 were collected by clinical monitoring of background characteristics and employment status of the intervention participants. Data of intervention participants in the period 2012-2016 were gathered in a prospective using self-report questionnaires. All measurements were performed at baseline (T0), post-intervention (T1, 1-year after baseline) and 2-years follow-up after baseline (T2). All over the years, the same operationalization of variables and measurement instruments were used.

With regard to the clinical monitoring, data on background characteristics, and status of paid and unpaid employment were prospectively registered at the reintegration company and outpatient rehabilitation clinics. For the present study these data were extracted from the clients' charts and entered in a digital data-extraction form by two researchers.

Outcome measures

In the intervention cohort, **paid employment** (primary outcome), and **unpaid employment** and **unemployment** (secondary outcomes) were assessed independently (0=no; 1=yes). Paid employment is defined as performing a paid job for at least 12h per week, in accordance with the definition of Statistics Netherlands at the time of this study [36, 37].

Background characteristics

Background characteristics were assessed at T0. In the intervention cohort, age, gender, onset of chronic physical condition (0 = acquired; 1 = congenital), special education (0 = no; 1 = yes), recipient of disability benefits (0 = no; 1 = yes), paid work experience (0 = no; 1 = yes), unpaid work experience (0 = no; 1 = yes), and job search period (0 = not yet; 1 = 0-1year; 2 = > 1 year) were recorded. Educational level was operationalized based upon the International Standard Classification of Education (ISCED): (0) low = pre-vocational practical education or lower (ISCED levels 1 or 2); (1) medium = pre-vocational theoretical education or upper secondary vocational education (ISCED levels 3 (vocational), 4 or 5); (2) high = general secondary education, higher professional education, or university (ISCED levels 3 (general), 6 or higher) [38, 39]. Severity of physical limitations was assessed using the 7-item indicator developed by Organization for Economic Cooperation and Development (OECD), assessing physical limitations (3 items) and limitations in hearing and seeing (4 items) on a 4-point Likert scale (1 = no physical limitations; 2 = slight physical limitations; 3 = moderate physical limitations; 4 = severe physical limitation). The severity of physical limitations was classified as severe if at least one

item scored 4, as moderate if at least one item scored 3, as slight if at least one item scored 2, and as no limitations if all items scored 1. This measure has a good reliability (Cronbach's $\alpha=0.79$) [40]. At baseline, problems related to competitive employment were identified using the Canadian Occupational Performance Measure (COPM) [41]. The COPM is designed to help patients identify important problems they experience in occupational performance in the domains of selfcare, productivity and leisure [42]. In a semi-structured interview a person selects a maximum of five prioritized activities that he or she wants, needs, or is expected to perform. Since we focused on problems related to competitive employment, we only report problems in the domains productivity and self-care (precondition for work). The COPM has good validity and interrater reliability, and is useful for young adults with physical disabilities [43].

External national reference data of young adults with chronic physical conditions

Data were collected by Statistics Netherlands, as part of a national health survey. In order to match the national reference data to the intervention cohort we used data of young adults a) with a self-report chronic physical condition, b) aged between 16 and 30 years; c) who indicated the severity of their physical limitations on the OESO-indicator [40]; and d) were recruited in the period 2007-2016. Each year Statistics Netherlands recruited a new population [37], and, in total, reference data of 2024 responders were used in the current study: 68.5% of them reported no physical limitations; 23.1% slight physical limitations; 4.6% moderate physical limitations; and 3.8% severe physical limitations. Responders of the reference data indicated whether they had a paid job for at least 12h per week, comparable with participants in the intervention cohort. Since the yearly employments rates in the period 2007 till 2016 and the number of respondents in a specific year were slightly differed between years, we calculated an averaged employment rate for the period 2007-2016, weighted for the number of respondents in a specific year. The averaged employment rate was also specified for four subgroups according to the severity of physical limitations.

Analysis

Descriptive statistics were used to describe the characteristics of the intervention cohort. Chi-square tests for dichotomous variables and independent t-tests for continuous variables were used in order to check for selective drop-out of responders at T1 and T2. Cochran's Q test served to test the overall development of intervention participants' employment rate (paid employment, unpaid employment and unemployment) over time. Post-hoc McNemar tests were used to compare paired proportions at two different assessment moments (0 to 1 yr; 0 to 2 yr; 1 to 2 yr). Similar analyses were conducted for subgroups regarding severity of physical limitations.

Chi-square statistics were applied to compare the paid employment rates of the intervention cohort at baseline and at 2-years follow-up to the national reference data and the effects were estimated by means of the relative risk ratios (RRs). For all analyses IBM SPSS Statistics version 21.0 (Chicago, USA) was used and the predefined level for statistical significance was 5%.

RESULTS

Background characteristics of the intervention cohort

Background characteristics of the intervention cohort ($n=90$) are presented in Table 1. Participants had a mean age of 24.5 years ($SD = 3.0$) at baseline, 14.4% had no physical limitations, 45.6% slight physical limitations, 20.0% moderate physical limitations and 20.0% severe physical limitations. Most of the participants had an acquired chronic condition (67.7%), a medium educational level (62.9%), were disability benefit recipient (75.6%), had prior paid work experience (77.6%) and/or unpaid work experience (64.9%). At baseline, most intervention participants reported problems with employment (77.8%), such as problems with finding a suitable job, writing an application letter, or doing a job interview. They also reported problems in preconditions for employment, such as housekeeping (50.0%), e.g. too tired after work to cook healthy food; functional mobility (38.9%), e.g. problem with transport to work or acquiring a driving license; personal care (31.5%), e.g. problems with dressing up in time; and education (9.3%), e.g. problems with finding a job that is consistent with their educational background or their education did not fit with their physical abilities.

Among the intervention cohort, nine participants could not be traced or did not respond for the assessment at post-intervention (T1), and twenty-six participants were lost at 2-years follow-up after baseline (T2). At T1, non-responders were more often male (88.9%; $p = 0.03$). Both at T1 and T2, non-responders were younger (mean age = 21.3 and 23.0 years respectively; $p < 0.001$ and $p = .01$ respectively). In addition, at T2 non-responders more often received disability benefits at baseline (64.0%; $p = 0.03$), had more prior paid work experience (63.7%; $p = 0.04$) and were less often searching for a job for more than one year (9.1%; $p = 0.01$). None of these characteristics correlated with the outcome measure paid and unpaid employment, and unemployment post-intervention and at two years follow-up.

Table 1. Background characteristics intervention cohort at baseline, post-intervention, and 2-years follow-up

	Study sample at baseline (N=90) [#]	Study sample at post- intervention (N=81) [#]	Study sample at 2-years follow-up (N=64) [#]
Age pre-intervention, mean (SD) [^]	24.5 (3.0)	24.9 (2.8) ^{**}	25.1 (2.7) ^{**}
Gender (male), n (%)	48 (53.3)	40 (49.4) [*]	32 (50.0)
Onset chronic condition (congenital), n (%)	30 (33.3)	27 (33.3)	22 (34.4)
Severity of the physical limitation (yes), n (%)			
No physical limitations	13 (14.4)	11 (13.6)	7 (10.9)
Slight physical limitations	41 (45.6)	38 (46.9)	32 (50.0)
Moderate physical limitations	18 (20.0)	14 (17.3)	10 (15.6)
Severe physical limitations	18 (20.0)	18 (22.2)	15 (23.4)
Educational level (yes), n (%) ^{^^}			
High	12 (13.5)	11 (13.8)	8 (12.5)
Middle	56 (62.9)	50 (62.5)	40 (62.5)
Low	21 (23.6)	19 (23.8)	16 (25.0)
Special secondary education (yes), n (%) ^{^^}	32 (36.0)	29 (36.3)	25 (39.1)
Disability benefit recipient, n (%) ^{^^^}	68 (75.6)	63 (81.8)	52 (85.2) [*]
Work status pre-intervention (yes), n (%)			
Paid	9 (10.0)	9 (11.1)	8 (12.5)
Unpaid	27 (30.0)	24 (29.6)	20 (21.3)
Work experiences pre-intervention (yes), n (%) ^{^^^^}			
Paid	38 (77.6)	30 (75.0)	24 (88.9) [*]
Unpaid	34 (64.9)	27 (67.5)	20 (74.1)
Job search period pre-intervention (yes), n (%) ^{^^^^}			
Not yet	9 (18.4)	6 (15.0)	2 (11.1)
0-1 year	24 (49.0)	19 (47.5)	10 (37.0)
> 1 year	16 (32.7)	15 (37.5)	14 (51.9) [*]
Reported problems related to competitive employment, n (%) ^{^^^^^}			
Productivity			
Paid or unpaid work	42 (77.8)		
Education	5 (9.3)		
Self-care			
Personal care	17 (31.5)		
Functional mobility	21 (38.9)		
Housekeeping	27 (50.0)		

[#]Unless otherwise indicated[^]n=88, n=80, n=63^{^^}n=89, n=80, n=64;^{^^^}n=86, n= 77, n=61;^{^^^^}n=49, n=40, n=27;^{^^^^^}n=54^{*}p < 0.05, ^{**}p < 0.01

Development of employment rates over time within the intervention cohort; and a comparison with external national reference data

Of the intervention cohort, nine participants (10.0%) had paid employment at baseline. Their jobs at that time, did not fit with their interests (n=1); educational level (n=5) e.g., student jobs or sheltered employment; physical capacities (n=2); or reason for work switch was unknown (n=1). All participants having paid employment post-intervention and at 2-years follow-up, found competitive employment, i.e. not sheltered. They were active in several sectors, for example ICT, healthcare, finance, retail, and education.

Overall, at baseline the paid employments rates of the intervention cohort were significantly lower compared to the national reference data of young adults with physical conditions (10.0% versus 52.9%, $p < 0.001$, RR = 0.19). Within the intervention cohort, paid employment significantly increased over time. The rates of paid employment were significantly higher at both post-intervention and 2-years follow-up (30.9%, 95%CI= 20.8-41.0; and 42.2%, 95%CI= 32.1-56.3 respectively) compared to baseline (10.0%, both p 's < 0.001). At 2-years follow-up the paid employment rate among the intervention participants (42.2%) approached the rate among the national reference data (52.9%, $p = 0.17$, RR = 0.80) (Table 2; Figure 1).

Unpaid employment rates increased from 30.0% at baseline to 35.8% post-intervention and thereafter decreased to 26.6% at 2-years follow-up (Table 2). However, this development over time was not significant ($p = 0.55$).

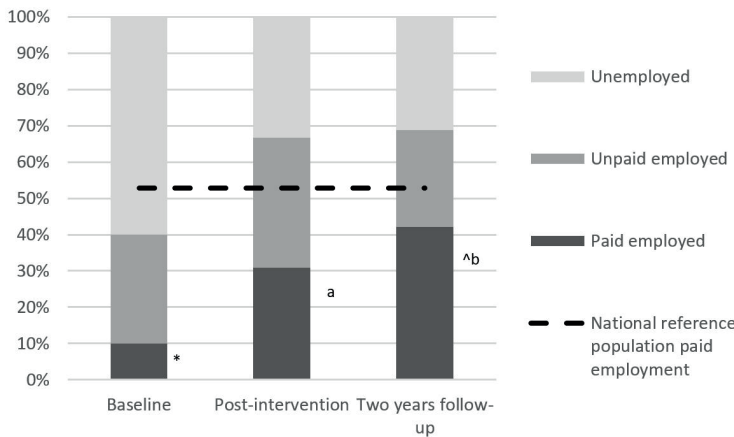


Figure 1. Development of paid employment rates within the intervention cohort, and a comparison with national reference data

[^] significant development over time within the intervention cohort
^{a, ^b} significantly differs from baseline within the intervention cohort
^{*} significantly differs from national reference data

Table 2. Employment rates within the intervention cohort, and a comparison with national reference data

	Intervention cohort				National reference data			
	Baseline		Post-intervention		2-years follow-up		Relative Risk ratio [#]	
	%		%	95% CI	%	95% CI	%	95% CI
Total								
(t0: n=90; t1:n=81; t2 =64)								
Paid	10.0 [*]	30.9 ^a	20.8-41.0	42.2 ^{ab}	32.1-56.3	52.9	50.9-54.9	.19
Unpaid	30.0	35.8		26.6				.080
Unemployed	60.0	33.3 ^a		31.2 ^{ab}				
No physical limitation								
(t0: n=13; t1:n=11; t2=7)								
Paid	23.1 [*]	36.4	8.0-64.8	42.9	6.2-79.6	56.0	53.6-58.4	.79
Unpaid	38.5	9.1		14.3				
Unemployed	38.5	55.5		42.9				
Slight physical limitation								
(t0: n=41; t1:n=38; t2=32)								
Paid	9.8 [*]	36.8 ^a	21.5-52.1	46.9 ^{ab}	29.6-64.2	50.6	46.4-54.8	.19
Unpaid	24.4	44.7		34.4				.93
Unemployed	65.9	18.4 ^a		12.7 ^{ab}				
Moderate physical limitation								
(t0: n=18; t1:n=14; t2=10)								
Paid	5.6 [*]	21.4	0.0-42.9	30.0	1.6-58.4	40.9	32.1-49.8	.14
Unpaid	27.8	21.4		30.0				.74
Unemployed	66.7	57.1		40.0				
Severe physical limitation								
(t0: n=18; t1:n=18; t2 =15)								
Paid	5.6 [*]	22.2	30.0-41.4	40.0 ^{ab}	15.2-64.8	29.2	20.1-38.3	.19
Unpaid	38.9	44.4		13.3				1.40
Unemployed	55.6	33.3		46.7				

[#]Relative risk ratio intervention cohort at baseline versus national reference data^{##}Relative risk ratio intervention cohort at 2-years follow-up versus national reference data^a significant development over time within the intervention cohort;^{a, b} significantly differs from baseline;^{*} significantly differs from national reference data

Subgroups analyses according to the severity of physical limitations

For all four severity subgroups within the intervention cohort (i.e. no, slight, moderate and severe physical limitations), the paid employment rates at baseline were significantly lower compared to the paid employment rates of the corresponding subgroups of the national reference data (respectively 23.1% versus 56.0%, $p = 0.02$, $RR = 0.79$; 9.8% versus 50.6%, $p = < 0.001$, $RR = 0.19$; 5.6% versus 40.9%, $p = 0.003$, $RR = 0.14$; and 5.6% versus 29.2%, $p = 0.03$, $RR = 0.19$).

Rates of paid employment in the intervention cohort increased over time in all severity subgroups, but this development was only significant in the subgroups slight and severe physical limitations ($p = 0.002$ and $p = 0.01$ respectively). The paid employment rate of participants with slight physical limitations significantly increased from 9.8% at baseline to 36.8% (95%CI = 21.5-52.1) post-intervention ($p = 0.03$), whereas, the employment the rates of participants with severe physical limitations significantly increased from 5.6% at baseline to 40.0% (95%CI = 15.2-64.8) at 2-years follow-up ($p = 0.03$) (Table 2; Figures 2b and 2d).

At 2-years follow-up, paid employment level of all subgroups of the intervention cohort approached the rates of the corresponding subgroups of national reference data (no physical limitation, $p = 0.37$, $RR = 0.79$; slight physical limitation, $p = 0.42$, $RR = 0.93$; moderate physical limitation, $p = 0.39$, $RR = 0.74$; severe physical limitation, $p = 0.28$, $RR = 1.40$) (Table 2; Figures 2a-2d).

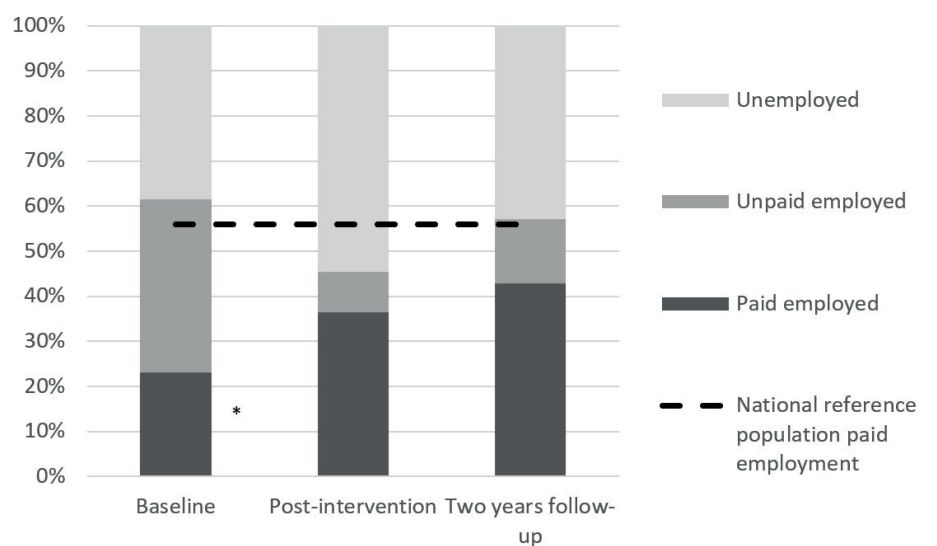


Figure 2a. Development of paid employment rates of subgroup no physical limitations within the intervention cohort, and a comparison with national reference data
* significantly differs from national reference data.

Increased employment rates after a vocational rehabilitation program

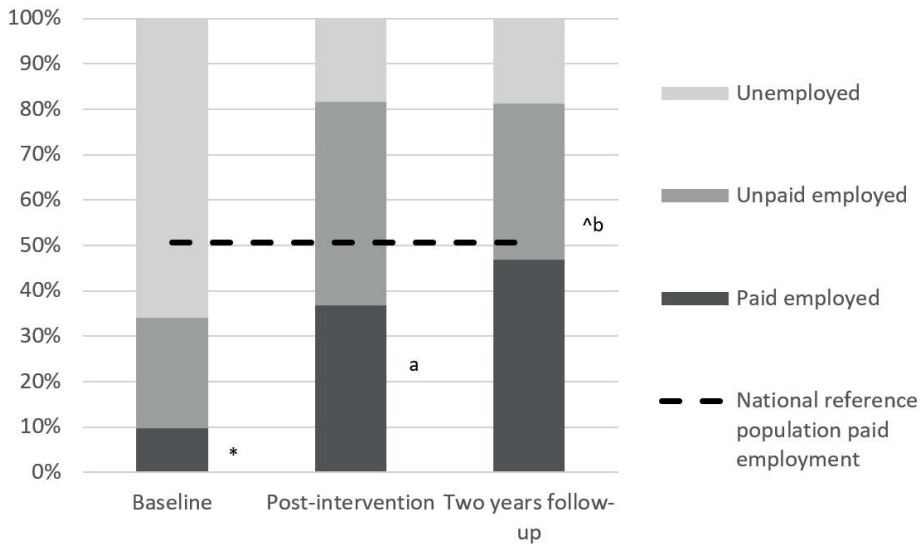


Figure 2b. Development of paid employment rates of subgroup slight physical limitations within the intervention cohort, and a comparison with national reference data

[^] significant development over time within the intervention cohort

^{a, b} significantly differs from baseline within the intervention cohort

^{*} significantly differs from national reference data

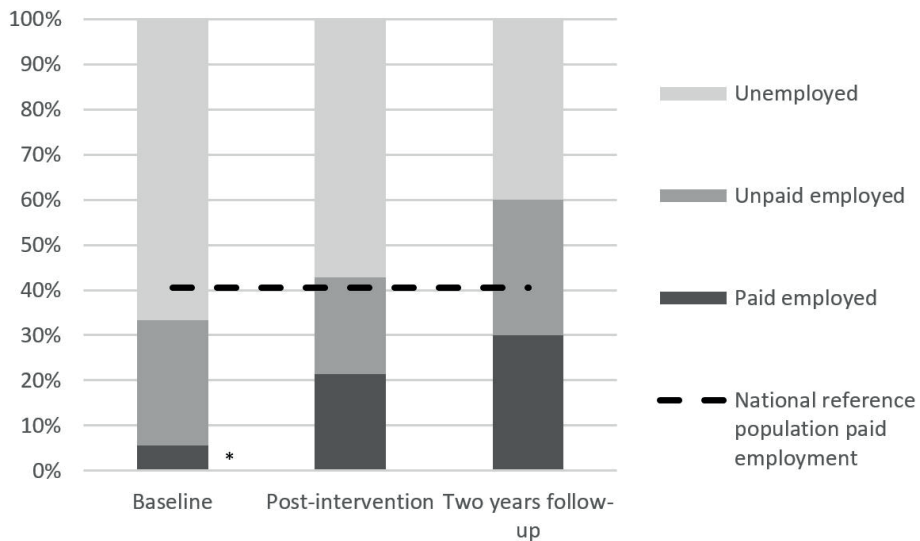


Figure 2c. Development of paid employment rates of subgroup moderate physical limitations within the intervention cohort, and a comparison with national reference data

^{*} significantly differs from national reference data

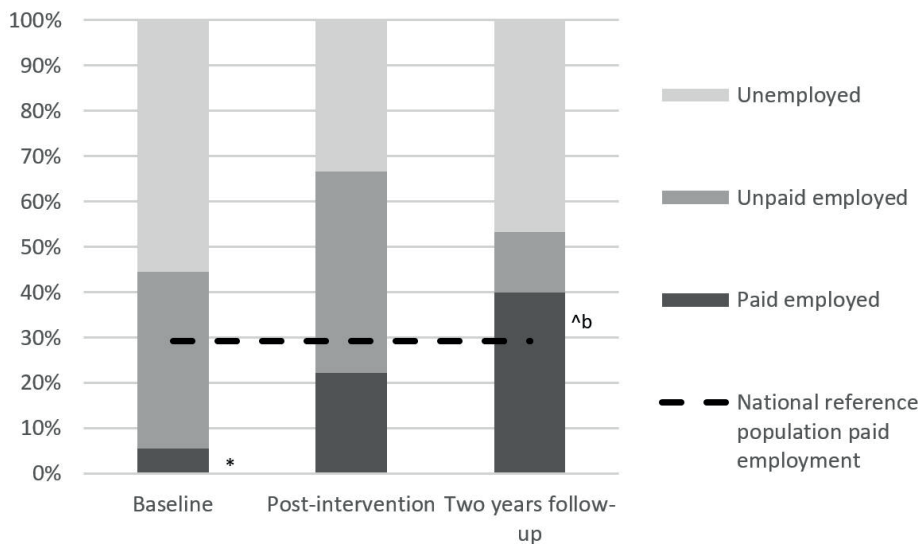


Figure 2d. Development of paid employment rates of subgroup severe physical limitations within the intervention cohort, and a comparison with national reference data

^a significant development over time within the intervention cohort

^b significantly differs from baseline within the intervention cohort

^c significantly differs from national reference data

DISCUSSION

In the present study we evaluated changes in young adults’ employment rates after participating in a vocational rehabilitation intervention. Starting from a disadvantaged position, the paid employment rate of the intervention cohort substantially increased on the short- and long term, and approached the employment rate of reference data of young adults with chronic physical conditions. In addition, intervention participants that achieved paid employment found a job in a competitive work environment. We evaluate these results as positive, considering the disadvantaged starting position of the intervention participants as compared to respondents of the national reference data. The reason is that the intervention cohort only included persons reporting problems in finding competitive employment, whereas the reference data also included persons who did not experience any employment problems, notwithstanding their chronic physical condition.

Most vocational programs for youth with disabilities as described in the literature are provided during adolescence, and facilitate the transition to temporary or seasonal employment [27-29]. Being engaged in education or doing temporal or seasonal work during adolescence is considered important, because it improves the psychosocial

development, by providing a context for developing life skills and exploring personal interests and abilities [11, 44]. However, previous studies showed that some young adults still lag behind in life skill development when finishing post-secondary education and they need more time to get ready for work [45, 46]. Some other authors of studies advised to provide this specific subgroup with tailored support [46], and the present results showed that 'At Work' may be an effective vocational rehabilitation intervention to fill this gap.

The 'At Work' intervention is, to our knowledge, among the first that is specifically developed for those young adults facing barriers to enter the competitive labor market. The program entails group sessions combining different components, such as peer-support, role-playing, and skills training, that are aimed to support life skills development, as well as job coach sessions aimed to support job placement and sustainable work participation. Results of previous studies showed that these components of the 'At Work' program, are perceived appropriate and effective to support life skill development of young adults [24-26, 28]. In addition, on-the-job training and job placement assistance were found to be effective for improving work participation of adults with chronic physical conditions [47, 48].

In our study we further specified intervention outcomes for subgroups with different levels of experienced physical limitations. We choose to estimate this by the OESO-indicator, since this was also available in the reference data. However, although the intervention cohort consisted of young people with chronic physical conditions who report problems with finding competitive employment, some of them reported no physical limitations according to the OESO-indicator. People in this subgroup are those without impairments in seeing, hearing or walking, but they still may have other physical impairments, for example chronic pain, that were not included in the indicator. Also, some of them might have other impairments in neuropsychological functioning which may influence their work participation, such as in attention, response inhibition, or memory, for example patients with an acquired brain injury [12]. Therefore, participants indicating no physical limitations on the OESO-indicator may still face problems when they enter the labor market or experience a negative impact of their chronic physical condition on their work participation. The question arises whether a classification based on severity of these specific *physical* limitations according to the OESO indicator is the most appropriate to get insight in intervention effects for specific subgroups with different levels of experienced impact of their chronic physical condition. It seems relevant to take other aspects, such as the participant's work readiness into account, when differentiating effects of vocational rehabilitation interventions between subgroups. However, these additional data were not available in the national reference data, and we had to use the

same definition of experienced impact of the chronic physical condition in both populations in order to allow comparison of the intervention cohort to national reference data.

Strengths and limitations

This is the first study, to our knowledge, that evaluates the effects of a vocational rehabilitation intervention on work participation in a large sample of young people with chronic physical conditions who experienced problems in finding competitive employment after finishing post-secondary education. Insights into effects of these types of interventions are important, since the work participation of young people with chronic physical conditions is known to lag behind compared to healthy age-mates [20, 21, 49].

The subgroup analyses entailed less power in each subgroup which may have led to trends that were not statistically significant, especially in the subgroups of no and moderately physical limitations. However, the subgroup analyses provided some additional information about intervention outcomes for persons with different severity levels regarding physical limitations in the present intervention cohort, that included a wide range of chronic physical conditions.

Since the 'At Work' program focused on a heterogeneous sample of young adults with a wide range of chronic physical conditions, it is not possible to relate findings to specific conditions. The heterogeneity of the population, however, is considered as a strong point of the intervention, since young adults with different chronic physical conditions may face similar challenges entering the labour market [50]. In fact, intervention participants recognized and learned from each other's experiences during the group sessions [30].

There is some discrepancy between the intervention cohort and the external national reference data regarding the type of chronic conditions. The intervention cohort consists of young adults with physical disabilities in rehabilitation care, whereas the reference data includes young adults with a broader range of chronic physical conditions, also addressing chronic conditions such as asthma or diabetes. Possibly, this difference might have influenced the estimated employment rates. Therefore it is worthwhile to directly compare the intervention cohort to a control group of young adults with similar chronic physical conditions, who are known in rehabilitation care but did not participate in the intervention.

CONCLUSION

Starting from a disadvantaged position, the level of paid work participation of the intervention cohort substantially increased on the short- and long term, and approached the level of work participation of national reference data of young adults with chronic physical conditions. The combination of group sessions and job coaching seems to be appropriate to support this specific group of young adults with chronic physical conditions who experience barriers in finding competitive and sustainable employment after finishing their post-secondary education.

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Declaration of interest

The authors have no competing interests to declare

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8

Effect evaluation of a vocational rehabilitation program for young adults with chronic physical conditions at risk for unemployment: A controlled clinical trial

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ABSTRACT

Background: The work participation of young adults with chronic physical conditions lag behind compared to healthy age-mates. 'At Work' is a vocational rehabilitation intervention provided by occupational therapists, that supports them for entering the competitive labour market after graduating post-secondary education.

Aim: To evaluate the effects of 'At Work' on self-efficacy, work-ability and employment status as compared to usual care.

Materials and methods: In total, 88 young adults were included in a multicentre controlled trial; 49 entered 'At Work', 39 received usual care. GEE-analyses were applied.

Results: Scores on all outcome measures substantially improved over time in the intervention group, but no significant effects were found as compared to the control group. The effect on general self-efficacy showed a positive trend in favour of the intervention group.

Conclusions and significance: Unlike previous study results pointing to positive outcomes of At Work, the current study did not support the effectiveness of this program on work-related self-efficacy, work-ability and paid employment, as compared to usual care. Yet, we did find an indication for positive intervention effect on general self-efficacy, which is known to be an important capacity to achieve social participation.

INTRODUCTION

Despite of having both the desire and ability to work, the work participation of young adults with chronic physical conditions lag behind compared to healthy age-mates [1-3]. Young adults with chronic physical conditions experience substantial difficulties when entering the competitive labour market, such as low self-esteem, lack of (life) skills, problems with disability disclosure, limited accessibility of buildings, limited adaptation of workplaces or working conditions, discrimination, and lack of support of manager or colleagues [4-10]. Yet, it is important to promote the work participation of these young adults, since it helps them to get control over their lives and fosters the development of self-identity and achievement of personal occupational goals [11-13].

Several vocational services are developed to support young people with chronic physical conditions in finding and maintaining employment. Occupational therapists are well-placed to deliver evidence based programs and services that promotes their work participation [14]. Most of the present interventions for this patient group mainly address the development of general life skills or provide support in finding temporary jobs as a student [15-19]. To our knowledge, none of these programs are primarily focused on supporting them to enter the competitive labour market after graduating from post-secondary education. To this specific aim, the 'At Work' program was developed in the Netherlands and implemented in some rehabilitation centres in the context of transition care [20-23]. 'At Work' is a vocational rehabilitation intervention provided by occupational therapists and jobcoaches and supports young adults with chronic physical conditions to find and maintaining suitable competitive employment [20-23]. The program is based on the Model of Human occupation (MOHO) [24] and has an empowerment perspective. MOHO is a commonly used model in occupational therapy. It states that persons generate and modify their occupations in interaction with the environment. The model discerns three main components: motivation, habituation and performance capacity. Accordingly, during the 'At Work' intervention motivation for employment, productive roles and routines, and work-related life skills are addressed. The program aims to enhance the self-efficacy at work, knowledge, self-awareness and skills of the individuals, in order to help them solve problems they face in their process to work. Self-efficacy refers to the capacity to take action and deal with life stressors [15]. 'At Work' offers a formal curriculum, including group education, peer-mentorship, simulation sessions, coaching and experimental learning [20-23]. These intervention elements are evaluated as useful and appropriate to improve young adults' (work-related) self-efficacy and life skills and to provide support for their vocational participation [15-19, 22, 25]. A feasibility study of the 'At Work' program showed an increase in paid employment rates from 8.3% to 33.3% after 1 year and to 41.7% after 2 years [20]. In a larger sample

we showed that – starting from a disadvantaged position – paid employment rates of former participants increased substantially over time, approaching employment rates of national data of young people with chronic physical conditions [23]. This was especially the case for those with severe physical limitations [23]. The present study aims to evaluate the program's effects on self-efficacy in solving work- and disease-related problems, self-perceived work-ability and employment status, as compared to usual care. We hypothesized a larger increase in work-related self-efficacy (primary outcome measure) in the intervention group (IG), compared to the control group (CG). The same pattern was expected for the secondary outcome measures addressing general self-efficacy, self-perceived work-ability and employment status. Thus, the study provides insights into the effects of vocational support provided by occupational therapists and jobcoaches.

MATERIALS AND METHODS

Design

We used the Consolidated Standards of Reporting Trials (CONSORT) [26] statement to structure this article. This study was designed as a non-randomized non-blinded multicentre controlled trial (Netherlands Trial Register NL3922/NTR4145) [27]. The IG consisted of participants of the 'At Work' program. The CG included young people with chronic physical conditions receiving usual rehabilitation care. All participants provided written consent and were entered in a lottery to win a tablet as token of appreciation. The Erasmus MC Medical Ethical Review Board approved the study (MEC 2012-381). All participating centres granted local approval.

Vocational rehabilitation program 'At Work'

'At Work' is an out-patient multidisciplinary vocational rehabilitation intervention for young people (16-27 years) with chronic physical conditions, aiming to foster (work-related) self-efficacy and life skills and, and to improve sustainable work participation. Participants must have finished post-secondary education, or do so within six months, and wish to enter competitive employment, i.e. not seasonal or temporary employment. According to the model of human occupation (MOHO) [24], motivation for employment, productive roles and routines, and skills are addressed during the 'At Work' intervention. In addition, an empowerment perspective is applied, in order to foster the self-efficacy and self-directed behaviour of participants [20-23].

Vocational services provided by jobcoaches of a reintegration agency are integrated with specialized rehabilitation care with a large share of occupational therapy. A rehabilita-

tion physician referred participants for the intervention, including six group-sessions and a one-year individual coaching trajectory. The intervention starts with an individual assessment by an occupational therapist. During this assessment, personal capacities are explored and personal goals for employment are determined using The Canadian Occupational Performance Measure (COPM) and the Occupational Performance History Interview (OPHI) [28,29]. After the individual intake, all participants take part in six group-sessions. These sessions are facilitated by an occupational therapist and a jobcoach, and a psychologist is involved in those group-sessions addressing mental issues. Group-sessions aim to empower the participants and, thus, increase their self-efficacy, knowledge, work and life skills, and awareness of their own values and needs. Participants are trained in solving work- and disease-related problems, to ask for modifications or adjustment of working conditions, and to disclose their condition to others. Group-sessions entail peer-support, role modelling, experimental learning and education; these intervention elements were evaluated as useful and appropriate to improve young adults' (work-related) life skills and to provide support for their vocational participation [15-19,22, 25]. The subsequent coaching trajectory is provided by the occupational therapist and a jobcoach. The occupational therapist help participants to reach personal goals as defined in the baseline assessment session that were not addressed during the group-sessions, e.g. to use public transport or to perform or organize their self-care activities. The individual coaching sessions provided by the jobcoach, are based on the supported employment methodology [30] and support finding and applying for a suitable job, and dealing with the social and physical work environment, i.e. informing the employer and colleagues about the chronic physical condition, arranging workplace modifications or adjusted working conditions. The content, frequency and duration of individual coaching were adjusted to the participants' personal needs and goals.

Study setting

'At Work' was developed in Rijndam Rehabilitation (Rotterdam), and subsequently implemented in two other rehabilitation centres, Reade Rehabilitation and Rheumatology (Amsterdam), and Basalt Rehabilitation (Leiden). These centres deployed an outpatient Young Adult Team, and collaborated locally with the same jobcoach agency 'VolZin'. Rehabilitation professionals in these centres completed a 'train the trainer course', consisting of two three-hour sessions, aiming to: a) educate about the theoretical base, content and practical organization of the intervention (including eligibility of participants, multidisciplinary collaboration); b) train interview skills for individual assessments at the start; c) inform and instruct professionals about this trial and data collection. Professionals indicated that the training gave them enough skills to implement and provide the 'At Work' Program. Also, engaging the same re-integration agency as part of the

intervention in all intervention centers, facilitated a comparable implementation of the intervention across centers. CG-participants were recruited in eight other rehabilitation centres, located in other regions of the Netherlands.

Participants and recruitment

In the IG, a rehabilitation physician referred young adults with chronic physical conditions for the intervention. These conditions include for example cerebral palsy, spina bifida, rheumatoid arthritis, (neuro)muscular disease, or acquired brain injury. Intervention participants were eligible for inclusion in this study if they met each of the following inclusion criteria: a) age 16-27 years; b) not employed, or not suitably employed, defined as employment not consistent with the individual's education, physical abilities and preferences; c) perceived problems with finding or maintaining competitive employment; d) finished education, or finishing education within 6 months; e) adequate understanding of Dutch language and; f) no severe intellectual impairments. A patient information letter was given to eligible patients.

To recruit participants for the CG, rehabilitation professionals (e.g. nurse, occupational or physical therapist) reviewed patient records in their databases, and compiled a list of candidates with chronic physical conditions who met the inclusion criteria. Employment status (inclusion criterion b) was checked by a self-report 4-item questionnaire enclosed with the patient information letter. Those who were not eligible for inclusion in the study, received an explanation for exclusion.

In order to detect a clinically relevant difference on the primary outcome measure Self-efficacy in solving work- and disease-related problems, with a power of 0.8 and alpha of 0.05, 86 participants (43 in each group) would be required. Recruitment was stopped when the predefined number of participants was reached.

Measurements

Procedure

For data collection we used the online application GemsTracker, which has been developed by Erasmus MC and partners for distribution of questionnaires during clinical healthcare research. GemsTracker automatically sent out questionnaires on the date of inclusion (baseline, T0) and one year later (post-intervention, T1). At two-year follow-up (T2), we contacted participants by phone to verify their employment status at that time.

Primary and secondary outcomes

The outcome measures for this study were selected in line with the main goals of the program, and address self-efficacy at work (primary outcome measure), and general

self-efficacy, as well as a person's work ability and employment status (performance) as secondary outcomes. The questionnaire Self-efficacy in solving work- and disease-related problems comprises 14 items, yielding a total score between 14 and 70 (Cronbach's $\alpha=.80$) [31]. The items of this questionnaire are presented in Box 1. We also assessed general self-efficacy with the 10-item General self-efficacy scale (GSES) as secondary outcome measure. The GSES yields a total score between 10 and 50 (Cronbach's $\alpha=.83$) [32]. In both measures a higher score indicates the favourable outcome. The Self-efficacy in solving work- and disease-related problems questionnaire was the best available measurement instrument for the present study, since it was validated for persons with chronic conditions in the Netherlands [31,33], and showed good internal consistency of the scales ($\alpha 0.8$) [31, 33]. Since the Self-efficacy in solving work- and disease-related problems questionnaire was only used in a limited number of studies so far [31,33] we additionally used the General self-efficacy scale (GSES), which is a valid measure that is broadly used in populations of young adults with chronic physical conditions [32, 34, 35].

Box 1. The items of the Self-efficacy in solving work- and disease-related problems questionnaire.

1. I am able to talk to my supervisor about adaptations in working conditions
2. I am able to mention problems that I experience at work
3. I am not able to set my boundaries at work
4. If I need some adjustments at work, then I am able to explain what I need
5. I am able to talk to my colleagues about my chronic condition
6. I am able to find out the work modifications that are available to me
7. I am able to figure out the legislation and regulations
8. I am able to ask the right people for help at work, if I can't do it by myself.
9. I am able to manage my work in such a way, that I do not get stressed
10. I am not able to negotiate with my supervisor about my tasks at work
11. If my colleagues react negatively to my chronic conditions, then I am able to deal with their reactions.
12. I am able to manage my work in such a way, that I have enough energy in the evening.
13. I am able to ask for help at work, if I can't do it on my own.
14. I am able to get the attention of my supervisor, when I have to tell something

Other secondary outcome measures addressed work ability and employment status, for which the instruments were in line with the previous feasibility study on the intervention [20]. Participants indicated their self-perceived work-ability on the Work Ability Index – Single item (WAS) [20, 36]. The WAS is derived from the WAI and consists of one question on the participant's self-reported current work ability compared to his highest work ability ever (scoring from 0 to 10) [36, 37]. Regarding employment status both paid and unpaid employment status (0=no; 1=yes) were specifically measured [38]. Paid employment status was defined as performing a paid job for ≥ 12 h/week, in accordance with the definition of Statistics Netherlands at the time of this study [39]. Furthermore, in the intervention group we also distinguished whether the paid employment was in a sheltered or competitive work setting. These secondary outcome measures were

validated for use or often used in studies on (young) people with chronic physical conditions [20, 36-38, 40].

Background characteristics

Background characteristics were assessed at T0. Age, gender, and onset of chronic condition (0=acquired; 1=congenital) were recorded. Severity of physical limitations was assessed using the 7-item indicator of the Organization for Economic Cooperation and Development (OESO) [41], assessing physical limitations (3 items) and limitations in hearing and seeing (4 items) on a 4-point ordinal score (1=no limitations (score of 1 on all items); 2=slight limitation (score of 2 on at least one item); 3=moderate limitation (scored of 3 on at least one item); 4=severe limitation (score of 4 on at least one item). Educational level was categorized according to International Standard Classification of Education ISCED: 0=pre-vocational practical education or lower; 1=pre-vocational theoretical education or upper secondary vocational education; 2=general secondary education, higher professional education, or university) [42]. We also recorded special education (0=no; 1=yes); having an established occupational disability (0=no; 1=yes); paid work experience pre-intervention (0=no; 1=yes); unpaid work experience at pre-intervention (0=no; 1=yes); and length of job search period pre-intervention (1=not yet searching; 2=0-1 year; 3= 1 year and over). At T1, using a custom-made questionnaire participants in the CG indicated the support for work participation they had received in the past year, and the setting in which the support was provided (1=jobcoach agencies, 2=rehabilitation care, 3=mental health care).

To monitor whether the 'At Work' intervention did not result in a decrease in health-related quality of life (HRQoL) due to physical or mental overload, in the IG HRQoL was measured using the 36-item Short-Form health survey (SF-36) [43]. For each domain of the SF-36, sum scores were calculated and transformed to a score of 0-100, with higher scores reflecting better HRQoL. In addition, scores were summarized in the Physical Component Summary (PCS) and Mental Component Summary (MCS), with an expected distribution of 50 (SD = 10) in the general population. The Dutch language version of the SF-36 is validated for use in populations with chronic conditions [44].

Analysis

Statistical analyses were performed with SPSS version 25.0. Demographic characteristics are presented using descriptive statistics. Chi-square-tests for dichotomous variables and one-sample t-tests for continuous variable were applied to test for differences between IG and CG at baseline, to check for selective drop-out of responders at T1, and for paid and unpaid employment also at T2.

General Estimation Equation (GEE) analyses with unstructured correlation structures were performed to determine intervention effects, following an intention-to-treat protocol. Imputation of missing data is not needed, because all available data of the outcomes are used and not only completed cases [45]. A GEE-model was chosen because a) it corrects for dependency of observations within an individual and for missing values; and b) it is robust for relatively small sample sizes [45]. To compare scores on self-efficacy in solving work- and disease-related problems, general self-efficacy and self-perceived work-ability for time interval T0-T1, the moment of measurement and the interaction between group allocation and moment of measurement were added to the GEE model as independent variables. For Paid employment status, which was assessed at T0, T1 and T2, group allocation, baseline values, measurement time and the interaction variable between group allocation and measurement time were added to the GEE model to estimate group differences for different time intervals (T0-T1 and T0-T2). For continuous variables, the regression coefficient (B) is displayed for the group variable (representing between-group differences). For dichotomous variables, between-group differences are indicated as odds ratios (ORs). The CG was specified as the reference group for all analyses. All models were adjusted for gender, onset of the chronic condition, severity of physical limitations, and having attended special education, because the IG and CG differed on these variables at T0.

Finally, in order to monitor changes in the HRQoL of the IG, we used descriptive statistics to check whether the HRQoL scores did not decrease during the intervention period in IG participants achieving paid employment at T1 (n=5), as compared to participants without paid work at T1 (n=24).

RESULTS

Study sample

Background characteristics of the total study sample, and broken down for the IG and CG, are presented in Table 1. In a 5 year period 49 eligible young adults who entered the 'At Work' program consented to participate in the study, of whom 30 (61.2%) completed the study at T1. In the same period 39 patients in the control centres consented to participate in the CG, of whom 28 (71.8%) completed the questionnaires at T1. The IG (n= 49) and CG (n=39) significantly differed for some background characteristics: participants in the IG were more often male (57.1% versus 35.9%), had less often a congenital condition (14.3% versus 43.6%) or severe physical limitations (14.3% versus 43.6%), and had less often attended special education (24.5% versus 46.2%).

Table 1. Background characteristics of intervention and control group

	Intervention group Study sample t0 (n=49) [#]	Control group Study sample t0 (n=39) [#]	Total Study sample t0 (n=88) [#]
Age, mean (SD)	23.38 (2.82) [^]	23.38 (2.55) ^{^^}	23.38 (2.69) ^{^^^}
Gender (male), n (%)	28 (57.1)	14 (35.9) [*]	42 (47.7)
Onset chronic condition (congenital), n (%)	11 (22.4)	17 (43.6) [*]	28 (31.8)
Severity of the physical limitation (yes), n (%)			
No physical limitations	4 (8.2)	2 (5.1)	6 (6.8)
Slightly physical limitations	23 (46.9)	13 (33.3)	36 (40.9)
Moderately physical limitations	15 (30.6)	7 (17.9)	22 (25.0)
Severe physical limitations	7 (14.3)	17 (43.6) ^a	24 (27.3)
Educational level (yes), n (%)			
High	10 (20.4)	16 (41.0)	26 (29.5)
Middle	33 (67.3)	18 (46.2)	51 (58.0)
Low	6 (12.2)	5 (12.8)	11 (12.5)
Special secondary education (yes), n (%)	12 (24.5)	18 (46.2) [*]	30 (34.1)
Established occupational disability, n (%)	32 (65.3)	31 (79.5)	63 (71.6)
Work experiences pre-intervention (yes), n (%)			
Paid	38 (77.6)	25 (64.1)	63 (71.6)
Unpaid	34 (69.4)	33 (84.6)	67 (76.1)
Job search period pre-intervention (yes), n (%)			
Not yet	9 (18.4)	8 (20.5)	17 (19.3)
0-1 year	24 (49.0)	20 (51.3)	44 (50.0)
≥ 1 year	16 (32.7)	11 (28.2)	27 (30.7)
Received vocational support past year pre-intervention			
Rehabilitation care (yes)	8 (16.3)	4 (10.3)	12 (13.6)
Vocational services (yes)	17 (34.7)	15 (38.5)	32 (36.4)
Mental care (yes)	1 (2.0)	1 (2.6)	2 (2.3)

Unless otherwise indicated

[^]=47; ^{^^} = 37; ^{^^^}n=84^{*} Significantly differs from intervention condition at T0 ($p < .05$)^a Adjusted residual = 3.1

Unfortunately, substantial drop-out occurred; the reasons for drop-out are unknown. At T1, drop-outs were younger than completers (respectively 21.46 yrs, SD=3.28 versus 23.73 yrs, SD = 2.43; $p = .004$), and at both T1 and T2, drop-outs were more often male (respectively, 78.6%, $p = .01$; and 62.9%, $p = .02$). At T2, drop-outs had more often experience with paid work at baseline (60%, $p = .04$) and were less often looking for employment for more than one year (17%, $p = .04$). None of these characteristics were significantly correlated with the primary outcome self-efficacy in solving work- and disease-related problems.

Support received in the CG

Completers in the CG ($n=28$) received usual care, which in 50% of the cases contained some form of vocational support, consisting of: a) support on work-readiness, e.g. gaining insight into personal interests, physical capacities, and mental capacities; assertiveness training; and education ($n=9$); b) job search assistance ($n=9$); or c) jobcoaching ($n=5$). Seven of them received two or more of these components of vocational support, for example support at work-readiness and job-search assistance or job-search assistance and jobcoaching.

Intervention effects

The data over time are shown in Table 2 and Figure 1 and Figure 2; the corresponding GEE analysis results are presented in Table 3. At T0, the IG started from a disadvantaged position as compared with the CG, with significantly lower scores on general self-efficacy ($p=.04$), self-perceived work-ability ($p<.001$), and paid employment ($p<.001$).

Table 2. Outcome measures for intervention and control groups

	Group	N	T0	T1	T2
		T0/T1/T2*	Mean ($\pm$ SD)/ Frequency (%)	Mean ($\pm$ SD)/ Frequency (%)	Mean ($\pm$ SD)/ Frequency
Primary outcome measure					
Self-efficacy in solving work- and disease-related problems (14-70)	IG	49/30	50.22 (7.26)	52.03 (8.23)	
	CG	39/28	52.95 (9.04)	53.57 (8.24)	
Secondary outcome measure					
General self-efficacy (10-40)	IG	49/30	28.86 (4.61)	30.63 (4.97)	
	CG	39/28	30.90 (4.54)	31.64 (5.41)	
Self-perceived work ability (1-10)	IG	49/30	5.86 (1.58)	6.23 (2.73)	
	CG	39/28	7.03 (1.71)	6.75 (1.76)	
Employment status					
Paid work >12 h/w (yes)	IG	49/40/27	1 (2.0)	12 (30.0)	13 (48.1)
	CG	39/34/26	11 (28.2)	17 (50.0)	17 (65.4)
Unpaid work (yes)	IG	49/40/27	15 (30.6)	14 (35.0)	6 (22.2)
	CG	39/34/26	15 (38.5)	8 (23.5)	4 (15.4)

* T2 data only available for paid and unpaid work

Self-efficacy in solving work- and disease-related problems, general self-efficacy and self-perceived work-ability

In both the IG and CG, participants improved on all outcomes during the intervention period. Corrected for gender, onset of the chronic condition, having severe physical limitations and having attended special education, no significant difference between IG and CG was found on the primary outcome self-efficacy in solving work- and disease-related problems ($b = .81$; 95%CI= -2.89 to 4.51; $p = .67$), nor on self-perceived work-ability ($b =$

Table 3. Longitudinal generalised equation results for between-group analyses in outcome measure, uncorrected and corrected for gender, onset of the chronic condition, severe physical limitations, and special education.

	Uncorrected		Corrected	
	B (95% CI)	P-value	B (95% CI)	P-value
Primary outcome measure				
Self-efficacy in solving work- and disease-related problems (14-70)	.13 (-3.52;3.79)	.943	.81 (-2.89; 4.51)	.667
Secondary outcome measure				
General self- efficacy (10-40)	1.78 (-.28; 3.84)	.091	2.02 (-.09; 4.12)	.061
Self-perceived work ability (1-10)	.14 (-.90; 1.18)	.794	.26 (-.77; 1.28)	.626
Employment status				
Paid work >12 h/w (yes)				
Δ T0-T1	.57 (.20; 1.61)	.288	.34 (.09; 1.25)	.104
Δ T1-T2	.67 (.22; 2.11)	.498	.50 (.10; 2.43)	.388
Unpaid work (yes)				
Δ T0-T1	.56 (.20; 1.59)	.275	1.77 (.54; 5.77)	.346
Δ T1-T2	.66 (.17; 2.65)	.562	1.35 (.35; 5.26)	.665

.14; 95%CI= -.90 to 1.18; $p = .63$). For general self-efficacy a trend was found ($b = 1.78$; 95%CI= -.28 to 3.84; $p = .06$), suggesting a larger increase in the IG as compared to the CG.

Employment status

In both the IC and the CG, the proportions of participants with paid employment increased during the intervention period and further increased up to 2-year follow-up. After correcting for gender, onset of the chronic condition, having severe physical limitations and having prior special education, no significant differences between IG and CG with respect to paid employment status or unpaid employment status were found after one year (T0-T1: respectively $b = .34$; 95%CI= .09 to 1.25; $p = .10$ and $b = 1.77$; 95%CI= .54 to 5.77; $p = .35$) and after two year (T0-T2: respectively $b = .50$; 95%CI= .10 to 2.43; $p = .39$ and $b = 1.35$; 95%CI= .35 to .53; $p = .67$).

Health related Quality of Life

The HRQoL scores in the IG were stable over time or seemed to increase more for participants with paid employment at T1 compared to those without paid employment at T1 (Appendix A).

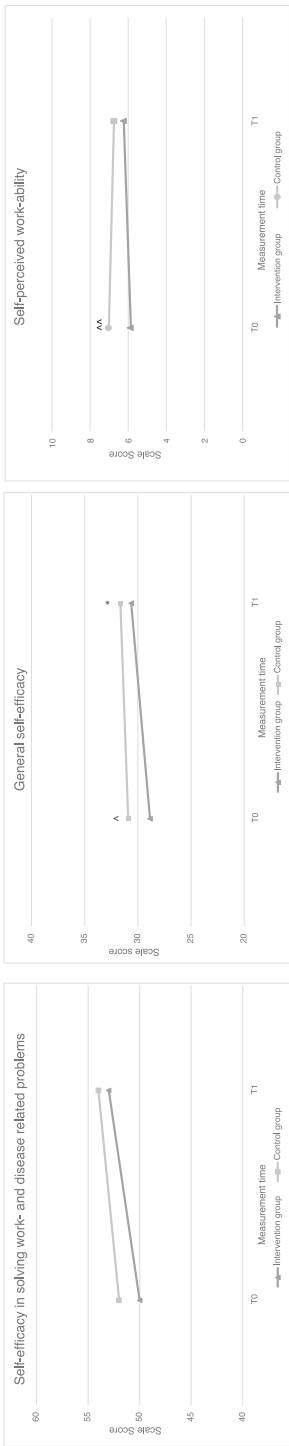


Figure 1. Self-efficacy in solving work- and disease related problems, general self-efficacy and c) self-perceived work-ability over time

Measurement time: T0=baseline, T1= post-intervention (at 1 year), T2= two-year follow-up;
Scale Scores on outcome measures: A) Self-efficacy in solving work- and disease-related problems questionnaire [Score range 14-70]; B) GSES, General self-efficacy scale [score range 10-50]; C) WAS, Single Work Ability Index – Single item [Score range 1-10].

^ Intervention group significantly differs from control group at T0, p=.04

* Positive trend was found, p=.06

^^ Intervention group significantly differs from control group at T0, p<.001

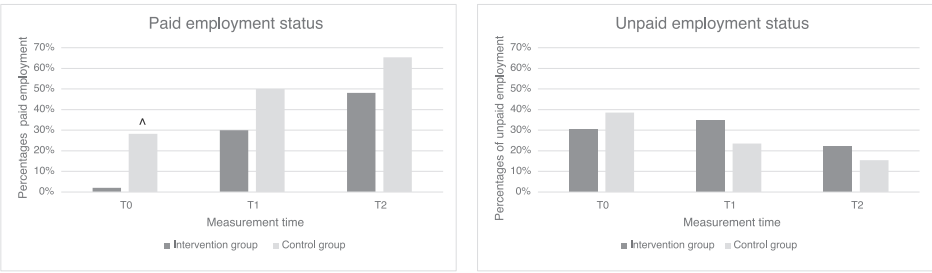


Figure 2. Development of paid and unpaid employment status over time
Measurement time: T0=baseline, T1= post-intervention (at 1 year), T2= two-year follow-up
^ Intervention group significantly differs from control group at T0, $p<.001$

DISCUSSION

The ‘At Work’ program is – to our knowledge – the first intervention that is specifically developed for young adults with chronic physical conditions facing barriers to enter the competitive labour market. In a previous study we showed that– starting from a disadvantaged position – paid employment rates of intervention participants substantially increased over time, approaching the employment rates of national reference data of persons with chronic physical conditions [23]. Results of a qualitative study indicated that young adults highly appreciated the components of the ‘At Work’ program (e.g. peer-support, skills training, role-playing) and perceived them effective to support their (work-related) self-efficacy and life skills development [22], which was in line with other studies [15-19]. Also for employed persons with chronic physical conditions hampering work participation on-the-job training and job placement assistance were found to be effective to improve their work participation [46-48]. In the present study we compared the effects of the intervention with a control group receiving usual rehabilitation care. In the IG all outcomes improved over time. In the CG, however, similar patterns were seen, albeit it to a lesser extent. As a result, this study did not provide evidence for the intervention’s effectiveness as compared to usual care. The results, however, do suggest a positive effect of the intervention on general self-efficacy, which tended to increase more strongly for participants of the ‘At Work’ program compared to those receiving usual care. Self-efficacy is considered important for youth success and being prepared for several aspects of adult life and independency [15].

Methodological considerations

A clear strength of this study is the two-year follow-up for the outcome paid employment status. This met the expectation that young adults with chronic physical disabilities have to overcome many obstacles to achieve paid employment (e.g. low self-esteem or a lack of life skills, problems with disability disclosure and travelling to work), resulting in

longer lead times before effects of an intervention on this outcome can be detected [20]. Also, the long-term follow-up was substantiated by the increasing employment rates of former 'At Work' participants until 2 years after the intervention. [23].

Also, some weaknesses should be mentioned. First, the recruitment of participants for the CG in this non-randomized design was difficult and might have resulted in a less comparable CG. In contrast to the intervention centres, most control centres did not provide specialized transition care, and the continuation of rehabilitation care is often hampered between paediatric and adult care. Thus, the control centres might have lost contact with young people in their early twenties with specific needs regarding social participation, including employment, and had to rely on former patient files for the inclusion. The flawed comparability of both groups might have hampered the detection of differences in intervention effects between the two groups. At baseline, the work outcomes of the IG and CG differed substantially, with higher scores on all outcomes in the CG, indicating less severe problems on average or different problems (e.g. overestimation of own abilities). Also, half of the CG did receive some kind of vocational support, which was probably adequate for them to achieve positive outcomes. It is important to further investigate what works for whom, and more specifically which young adults have a need for more intensive vocational support, such as the 'At Work' program. Especially occupational therapists can, based on their clinical experience, help to identify subgroups of young adults with specific support needs [14]. These insights may help to further tailor the intensity of vocational support to a person's needs, and to fine-tune the inclusion criteria for the more intensive 'At Work' program.

Other limitations of this study address the outcome measures used. For work-related self-efficacy we had to use a less-than-ideal measurement instrument for this specific target group that is at the start of a work career, a situation which is inherently different from dropping-out and returning to work. A sensitive outcome measure specifically addressing work-related self-efficacy or work readiness of young adults entering the labour market was not yet available. We selected the 'self-efficacy in solving work- and disease-related problems questionnaire' as primary outcome measure, although this was originally developed for *adult employees* with a chronic physical disease, who experienced problems in their employment [31]. This instrument showed to be less appropriate to measure work-related self-efficacy among *young* adults with ample work experience and a lack of context to appraise their self-efficacy at work. A possible indication for this problem is a relatively low Cronbach's alpha for this measure in the present sample (.67). There is a need for age- or developmentally appropriate outcome measures to capture work-related self-efficacy or work readiness in starters entering the labour market. Furthermore, whereas we know from the measurements that paid em-

ployment of participants in the IG was all competitive, we assume that the assessed paid employment in the CG also comprised sheltered employment. The latter assumption is based on the observation of relatively high proportions of CG-participants with severe physical limitations and previous special education (Table 1). We estimate that about 15% of the paid employment of participants in the CG may refer to sheltered employment, based on the work participation of young adults with cerebral palsy [49]. In future studies on the effects of vocational rehabilitation interventions, it is recommended to distinguish between sheltered employment and regular employment in the assessment of employment status.

Clinical implications

Starting from a disadvantaged position, at least equivalent effects were found in the intervention group as compared to a control group in rehabilitation care. With that and because of the effects of the 'At Work' program on *sustainable* work participation in the long term, it might be worth providing such an intensive program to support young adults with chronic physical conditions who face barriers as starters in the competitive labour market. The results suggest to further tailor the type and intensity of vocational support for young adults with chronic physical conditions entering the labour market.

CONCLUSION

Unlike several previous study results pointing to positive outcomes of the 'At Work' program, the current study did not add to the evidence on its effectiveness on work-related self-efficacy, self-perceived work-ability and employment status as compared to usual care. Yet, we did find an indication for positive intervention effect on general self-efficacy, which is known to be an important capacity to achieve optimal social participation. Challenges to optimize future effectiveness studies on vocational rehabilitation programs in this age group transitioning to adulthood address the comparability of the intervention and control group and the use of a more sensitive outcome measure to capture preparedness for work among starters in the labour market.

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Declaration of interest

The authors have no competing interests to declare.

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Appendix A: Health-related quality of life within intervention group

	Paid work at T1	N T0/T1	T0 Mean ($\pm$ SD)	T1 Mean ($\pm$ SD)
Health-related quality of life (1-100)				
Physical Functioning	Yes	12/5	60.42 (24.81)	72.00 (32.90)
	No	28/24	59.46 (26.29)	60.00 (26.46)
Role limitation due to physical problems	Yes	12/5	58.33 (38.92)	65.00 (48.73)
	No	28/24	36.61 (41.10)	26.04 (34.95)
Bodily Pain	Yes	12/5	74.67 (26.05)	84.80 (20.81)
	No	28/24	60.04 (27.44)	53.46 (24.01)
Energy Vitality	Yes	12/5	56.67 (20.15)	69.00 (14.75)
	No	28/24	49.11 (21.00)	47.08 (22.16)
General Health perception	Yes	12/5	56.33 (20.78)	69.60 (16.40)
	No	28/24	52.79 (21.39)	48.63 (27.21)
Social functioning	Yes	12/5	71.88 (21.40)	77.50 (22.36)
	No	28/24	58.48 (25.92)	60.94 (22.52)
Role limitation due to emotional problems	Yes	12/5	72.22 (39.78)	80.00 (44.72)
	No	28/24	73.81 (39.91)	51.39 (45.02)
Mental Health	Yes	12/5	66.67 (16.74)	72.00 (10.20)
	No	28/24	69.00 (15.11)	63.67 (18.61)
Physical component summary	Yes	12/5	25.40 (1.23)	26.18 (1.67)
	No	28/24	24.90 (1.54)	24.88 (1.55)
Mental component summery	Yes	12/5	23.55 (1.57)	23.97 (.69)
	No	28/24	23.53 (1.59)	23.10 (1.95)

9

General discussion

INTRODUCTION

The studies presented in this thesis focused on professional support to young people growing up with chronic physical conditions, aimed to develop their self-management skills and to achieve vocational participation. In this concluding chapter, the main findings are discussed in the context of available knowledge about self-management support and support for vocational participation of young people with chronic physical conditions. In addition, methodological considerations are discussed as well as implications for further practice and research.

MAIN FINDINGS

Part 1: Self-management support

The first part of this thesis deals with the content, effectiveness and effective components of timely self-management support for young people with chronic physical conditions. Moreover, we aimed to define a subgroup who might need timely psychosocial support – i.e., support to develop self-management skills and to achieve (pre)-vocational participation.

In a systematic review, we found that existing self-management interventions were predominantly focused on the medical domain, considering the medical aspects of the chronic condition. Possible psychosocial challenges often remained unaddressed (Chapter 2). In line with these findings, we found some evidence for effectiveness of self-management interventions on outcomes such as disease knowledge and treatment adherence, but we found conflicting evidence on improvement of one's ability to deal with a chronic condition in daily life, of psychological outcomes, and of quality of life (Chapter 3). Online peer-support formats, such as Skype video calls, e-mentoring mobile apps or interactive websites, were found to be effective components of self-management interventions that aimed to improve one's ability to deal with a chronic condition in daily life. Group sessions seemed to be an effective format for self-management interventions that focused on improving psychosocial outcomes (Chapter 3).

In the Netherlands, young adults with a childhood onset of a chronic condition could apply for a disability benefit (Wajong) when they experience problems in vocational participation. Young adults who qualified for a benefit reported a greater extent of physical disability, attended less often special education, experienced a greater impact of the chronic condition on school and vocational functioning, and reported a lower health-related quality of life during adolescence in comparison to those who did not ap-

ply. Those adolescents may benefit from timely support for improvement of self-management skills, life skills and (pre-) vocational participation (Chapter 4), to counteract a substantial delay in psychosocial development and a low health-related quality of life.

Part two: Vocational support

Part two of this thesis addresses outcomes of vocational rehabilitation programs in general and of the vocational rehabilitation program ‘At Work’ in particular.

We performed a systematic literature review on vocational rehabilitation interventions for persons with chronic physical conditions, which showed positive effects on work status (i.e., return to work, or employment rate) and work attitude (i.e., work readiness, employment activities, employability or self-efficacy at work), but not on work productivity (i.e., hours per week worked or duration of sick leave). The content and characteristics of these interventions varied widely. Effective interventions consisted of multiple components, whereas other content or delivery characteristics were not specifically associated with positive outcomes (Chapter 5).

‘At Work’ is among the first vocational rehabilitation programs designed for young adults (16-27 years) with chronic physical conditions who have difficulty in finding competitive employment after finishing post-secondary education. It starts with six group sessions, followed by a one-year coaching trajectory. An innovative aspect of this program is the comprehensive approach integrating rehabilitation care provided by an occupational therapist and vocational services offered by a job coach of a re-integration agency. A detailed description of the ‘At Work’ program is presented in Chapter 6.

The results of the studies on the ‘At Work’ program were promising. A qualitative study revealed that participants perceived peer-support, the job-interview training, and job placement as beneficial attributes of the program, since these offered opportunities to experience challenging real-world situations that provide new insights, a higher self-efficacy and improved life skills. Moreover, the adjusted work conditions and coaching of the work supervisor helped participants to overcome barriers to competitive employment, such as limited physical capacities and negative attitude of the employer. Recommendations for improvement reported by the participants included additional training on assertiveness and communication skills regarding disability disclosure, which could equip them with the necessary social and communication skills to deal with the work environment (Chapter 6).

Quantitative studies revealed that the proportion of ‘having paid work’ increased substantially among the ‘At Work’ participants (n=90), who had started from a very

disadvantaged labor market position. Two years after the start of the 'At Work' intervention, their level of work participation approached the level among young adults with chronic physical conditions recognized in national reference data (Chapter 7). This was especially the case for young adults with severe physical limitations.

In a controlled clinical trial, we directly compared the outcomes of the 'At Work' intervention group to a control group of young adults with similar chronic physical conditions who received usual rehabilitation care, including usual vocational interventions (Chapter 8). Again, 'At Work' participants started from a very disadvantaged position, but their self-efficacy in solving work- and disease-related problems, general self-efficacy, self-perceived work-ability and paid employment increased substantially over time. Participants in the control group showed these achievements as well, although it seemed to a lesser extent. Still, the improvement of general self-efficacy among participants of the 'At Work' intervention tended to be higher than that of the participants in the control group.

GENERAL DISCUSSION

Role management within self-management support for young people with chronic physical conditions

Advancements in the treatment of chronic or life-threatening conditions have resulted in increasing numbers of young people with chronic physical conditions surviving into adulthood [1-2]. The transition to an independent adult life requires complicated life changes for every young person, which may be more difficult for young people with chronic physical conditions compared to healthy peers. Some of them successfully integrate into adult life: they start intimate relationships, leave home, follow education and find employment. However, many others do not succeed in all these life areas [3-8]. Moreover, at a certain age they are expected to take charge on their medical management, but this do not always start naturally [9, 10]. It seems necessary, therefore, to support the development of self-management skills [11-13].

According to the definition of self-management and self-management support proposed by Barlow, self-management include three elements: medical management (considering the medical treatment), role management (considering participation in society), and emotion management (considering well-being)" [14, 15]. In a literature review, we showed that transitional care is still mostly focused on medical management issues, with less attention to emotional and social aspects of growing up with a chronic condition (Chapter 2). The three different self-management domains as defined by

Barlow [14,15], are, however, interrelated. Thus, a focus on the medical management may positively affect the non-medical outcomes as well. A better understanding of the chronic condition and its treatment helps a young person to integrate the chronic condition in daily life and be more confident in managing the condition. Therefore, role and emotional management should be considered in self-management support as well.

Our study on predictors of a delay in psychosocial development indicates that self-management support for specific subgroups of adolescents with chronic physical conditions needs to be timely introduced (Chapter 4). The transition to adulthood is a stepwise process rather than an event, in which young people must reach several developmental milestones and acquire age-appropriate life skills at each step [5, 17-19]. Adolescence and young adulthood are key phases for people with chronic physical conditions to develop these skills and to engage in social roles toward independence [20-22]. Therefore, it is important to substantially invest in psychosocial development and (pre)vocational participation [7, 23, 24]. The recently published Netherlands Transitional Care quality guideline [*Kwaliteitsstandaard Transitiezorg*] underlines the importance of transitional care and self-management support for young people with chronic physical conditions. This guideline mainly addresses medical aspects of a chronic condition, while still paying limited attention to social participation. Recommendations on supporting the psychosocial development of young people with chronic conditions must be incorporated in more detail in future versions of this guideline. Individual transition plans, for instance, help practitioners to monitor these young people's psychosocial development and to open up discussions in the consultation room about developmental tasks and possible challenges [26, 27]. The Skills for Growing Up tool and the Ready Steady Go tool are examples of tools that facilitate the communication about the development of age-appropriate life skills [28]. The topics addressed can serve as a basis to decide what actions on medical management and psychosocial development must be taken, and support programs can be started. These support programs aim at fostering psychosocial development based on the positive youth development theory [29]. According to this theory, interventions should incorporate the 'Big Three' principles: a) incorporating mentorship roles; b) applying a focus on the development of life skills; and c) facilitating sustained and supportive youth-adult relationships (adult role models) [29]. In our literature review on effective components of self-management interventions, we only found some evidence for the effectiveness of mentorship roles on dealing with the chronic condition in daily life (Chapter 3). However, intervention programs and outcome measures used in the evaluation studies were very heterogeneous, which downgraded the quality of evidence. More standardized research on the effectiveness of self-management interventions is needed.

Vocational support for young adults with chronic physical conditions

Young people with chronic physical conditions experience substantial difficulties in their vocational participation, such as problems with traveling to work, limited accessibility of buildings, limited adaptation of workplaces or working conditions, but also issues of disability disclosure, discrimination, lack of support of manager or colleagues, low self-esteem and inadequate work-related life skills [8, 30-36]. Initiatives and interventions at macro level (i.e.; national policies and legislation), meso level (i.e., local policies at municipalities, implementation agencies, and interventions within organizations) and micro level (i.e., access to dedicated vocational rehabilitation programs for people with chronic physical conditions), aim to intervene on these barriers and improve the vocational participation rate.

In the years that we performed the studies described in this thesis, young adults who were partially or fully incapable of working due to childhood onset chronic conditions were entitled to social benefit for young disabled persons under the Wajong Act (the Disablement Assistance Act for Handicapped Young Persons) [37]. The Wajong Act is described in more detail in the introduction paragraph of this thesis. The disability benefit consisted of (supplementary) income support, and support in finding and sustaining employment for those who have some work ability [37, 38]. In 2015, new legislation was implemented in the Netherlands with the so-called 'Participation Law' [*participatiewet*]. Under this law, all people with chronic conditions who have some work ability and who have a need for vocational support are entitled to a benefit that only includes support in finding and sustaining employment. Those people were listed at the target group registers [*doelgroepenregisters*]. People with chronic conditions who live independently without a partner, are entitled to some income support under the scheme of social assistance benefit [*bijstandsuitkering*]. Companies are obligated to arrange jobs for people listed at the target group registers, according to the job agreement law [*Wet banenafspraak*], which is part of the Participation Law. After the implementation of the Participation Law, people listed at the target group registers have more often paid work as compared to people who applied for a Wajong benefit. However, many of them have only temporary jobs [39].

The Participation Law shifted the responsibility for providing vocational support for young adults with chronic physical or mental conditions from The Dutch National Employee Insurance Agency (UWV) to the 345 municipalities. Many municipalities have currently limited insight in the subgroup of people with chronic physical conditions [39]. Financial resources for vocational support have been reduced, and some subgroups of people with a severe physical limitation or relatively low work ability are nowadays not supported under this legislation, because in their regions the social service was

judged as too expensive for these target groups [39]. In line with this policy change, we noticed that the referral of young adults with chronic physical conditions to the 'At Work' intervention was severely hampered after the implementation of the Participation Law, and consequently the 'At Work' program can nowadays no longer be provided. Yet, research showed that it is very important to support their personal development, self-management and vocational participation, making use of specialized interventions that help them to get control over their lives, improve self-management and work readiness and foster the development of self-identity and achievement of personal occupational goals [11, 17, 23, 29, 40]. The use of tailored vocational interventions may result in a positive return on investment on a macro level, especially for the members of the target group who aspire to enter the competitive labor market. We have found indeed that a relatively high proportion of participants of the 'At Work' program (42.4%) had realized paid employment two years after the start of this intervention. Sustainable employment can – in the long term – result in fewer disability claims and higher quality of life [41, 42].

Employers, too, are important actors in stimulating the work participation of people with disabilities. As indicated in our study, young people with chronic physical conditions are often confronted with a negative attitude of an employer, which hampered their work participation. It is important to intervene on this negative attitude and to encourage and support employers in hiring people with disabilities. Vocational interventions with a job coaching component, like the 'At Work' program, can generate jobs for young people with disabilities, which otherwise would not have been created because of employers' reticence to hire people with disabilities [39, 43].

At micro level, it is important to support young people with chronic physical conditions to enter the (competitive) labour market. From our literature review regarding the effectiveness of vocational rehabilitation programs for people with chronic physical conditions, we concluded that, in general, it is beneficial to apply targeted interventions, irrespective of the precise diagnoses or intervention characteristics (Chapter 5). On-the-job training and job placement assistance seemed particularly effective in improving work participation of people with chronic physical conditions [44, 45]. Multiple component interventions seemed to be more effective in improving work participation than single component interventions (Chapter 5). To our knowledge, in the Netherlands, three initiatives other than the 'At Work' program, support vocational participation among young adults with a chronic physical conditions, i.e. 'Emma at work' [46], 'Brains4U' [47], and The Class [48]. The latter two initiatives are specially focused on young adults with acquired brain injury. Systematic evaluation of the Class program, showed that 50% of the participants had paid or unpaid work one year after that start of the intervention.

A vocational rehabilitation intervention for a specific age group: ‘At Work’

Conclusions about the effects of the ‘At Work’ program

Based on the series of studies we concluded that the results of the ‘At Work’ program are promising, since the paid employment rates of ‘At Work’ participants substantially increased in the short and long term (31% and 42%), and approached the paid employment rates found in national reference data (53%). Especially young adults with severe physical disabilities – which group showed the largest improvement of employment rates – seemed to benefit from the ‘At Work’ intervention. In a controlled trial, however, we did not find evidence that ‘At Work’ was more effective than usual care, except for a trend that the ‘At Work’ participants increased more on their general self-efficacy. Since the ‘At Work’ program is the first intervention specifically developed for young adults on the verge of entering the (*competitive*) labor market *after finishing their post-secondary education*, it is not possible to compare the effects of the ‘At Work’ program with other vocational rehabilitation interventions for this specific age group. Nevertheless, we know from adjacent literature that the components of the ‘At Work’ program, such as role-playing opportunities and peer-support during group sessions, job-search assistance and coaching on the job, can be effective to foster life skills development of adolescents with chronic physical conditions and to support their work participation [44,45-51-54].

Perceived effective components of the ‘At Work’ program

‘At Work’ participants mentioned that especially their life skills were trained during the group sessions. They considered peer-support and the job-interview training as beneficial attributes, as these provide them with respectively self-efficacy and communication skills for disability disclosure. They also recommended including more role-playing opportunities in order to stimulate assertiveness and communication skills for disability disclosure. Assertiveness is an important life skill for arranging adjusted work conditions or asking for help at work, if necessary. Accordingly, vocational rehabilitation for young people with chronic physical conditions should be focused on personal abilities, knowledge, (life) skills, and attitude. Actually experiencing challenging real-world opportunities for vocational participation, role playing opportunities and peer-support during adolescence and young adulthood, will bring new insights, and improve self-efficacy and life skills [7, 20-23, 40]. Mastering life skills may help overcome possible barriers in a person’s vocational participation [5, 7, 19].

Participants of the ‘At Work’ program also found the job placement assistance helpful. During the individual coaching trajectories, the job coach arranges adjusted work conditions and informs supervisors at the workplace about the chronic condition and the

young adult's work abilities. This approach has been shown to be effective to improve suitable employment among people with chronic conditions [45], which was also confirmed by 'At Work'. Nevertheless, it might also be important to further train participants' life skills and to empower them during their individual trajectories, thus enabling them to take more control over arranging their work conditions [40]. In the process leading to sustainable employment, situations may arise that are perceived as difficult to deal with. These situations are real-world opportunities to enhance the life skills acquired through the individual job-coaching intervention component.

IMPLICATIONS FOR CLINICAL PRACTICE

Part 1: Self-management support

The results of this part underline the importance of a lifespan approach throughout pediatric rehabilitation care and the transition to adult care. The transition phase should include support on psychosocial aspects of functioning with a chronic condition as well as on medical aspects. This support should be provided by specialized and dedicated teams of healthcare professionals. Rehabilitation professionals of the Young Adult Teams have the task to monitor their clients' psychosocial development, for which standard instruments such as the Rotterdam Transition Profile, Skills for Growing up tool or the Ready Steady Go tool can be used. These instruments help the health professionals to gain insight in a person's level of functioning in the various participation domains. Using one of these instruments reduces the risk that some participation domains, such as work participation, are not addressed during consultations. If necessary, rehabilitation professionals should refer a client to individual or group-based interventions provided by occupational therapists, psychologists or social workers, that are aimed at single or multiple participation domains. Especially young people who attended mainstream education and those with severe physical disabilities are vulnerable for a delay in psychosocial development (Chapter 2). We recommended, therefore, inviting them frequently for consultations to discuss their reported outcomes and to monitor their psychosocial development. If needed, timely psychosocial support can be offered, with attention to actual or future work participation.

Part 2: Vocational support

The 'At Work' program is a promising intervention to support the vocational participation of young adults with chronic physical conditions who experience problems with finding and maintaining competitive employment after finishing their post-secondary education. Especially young adults with severe physical disabilities seem to benefit from the 'At Work' program. Unfortunately, the implementation of the Participation Law implied

that much fewer young adults could attend the 'At Work' program. As a consequence, this intervention can no longer be provided. An evaluation of the implementation of the 'Participation Law' showed that municipalities still have a limited view of the target group of young people with chronic physical conditions, especially those in mainstream education [39]. From our studies on the 'At Work' intervention, some lessons can be learned for future vocational rehabilitation interventions for young adults with chronic physical conditions:

- a) In order to improve their work readiness after finishing post-secondary education, it is important to train work-related life skills;
- b) Appropriate elements are role-playing opportunities– i.e., job interview training, strategies to disclose the chronic condition at work and asking to adjust working conditions – and peer-support during group sessions;
- c) job coaching, job carving and job crafting are suitable strategies to support work participation.
- d) The job coaching itself might be improved when the job coach can directly intervene in situations that a young adult perceives as difficult to deal with, instead of a few days later during a planned coaching session [52].
- e) The use of online peer- or job coach supporting formats, such as Skype video calls, e-mentoring mobile apps or interactive websites, might be suitable to incorporate into the jobcoaching, as indicated in our review. A recently described e-mentoring program 'Empowering youth towards employment' is an example of a promising intervention for youth with physical disabilities to prepare for employment [53]. However, in the Netherlands job coach interventions are not covered by the Health Insurance Act, and thus are not part of rehabilitation care. These interventions have to be provided by municipalities, but these may lack the connection with vocational rehabilitation care.
- f) Lastly, more support to employers is needed, aimed at encouraging their willingness to facilitate the vocational participation of persons with chronic physical disabilities.

METHODOLOGICAL CONSIDERATIONS

Part 1

The content and intervention elements of self-management support interventions differed across studies. Moreover, the outcome measures in the effectiveness studies varied. Therefore, results about effective intervention components have to be interpreted with some caution, since the quality of evidence of the studies was low.

Part 2

The 'At Work' program is the first intervention specifically developed for young adults with chronic physical conditions who face barriers to enter the competitive labor market. Also, to our knowledge, ours is the first study on the effectiveness of vocational rehabilitation interventions for this target group in a *controlled clinical trial*. Consequently, it is not possible to compare results of the 'At Work' program with those of other vocational rehabilitation interventions for this target group. In addition, the social context – i.e., the social legislation and regulations, national disability benefits and the national employment opportunities – differs widely across countries. This hampers comparison between countries and their effects. Moreover, the social context may have changed over years, which complicates a meta-analysis of the effects of vocational rehabilitation interventions.

We were confronted with quite some barriers to performing a controlled clinical trial in this specific target group. First, it was very hard to include a control group that was comparable with the intervention group. Rehabilitation centers that invited participants for the control group differed on the transitional care they offered. As a result, at these centers, young adults' needs for vocational support were not systematically discussed during consultations or registered at the client charts, which hampered their identification in the inclusion process.

A second barrier regarded the selection of a valid outcome measure addressing work-related self-efficacy or work readiness that was suitable and preferably validated for young adults without former employment experience. After our search, we decided to use the 'self-efficacy for solving disease and work-related problems questionnaire', which originally had been developed to measure work-related self-efficacy in *adult employees* with a chronic condition who experienced problems in competitive employment [54]. With the present knowledge, we now conclude that this instrument was hardly appropriate to measure changes in work-related self-efficacy in *young* adults with chronic physical conditions, who (mostly) do not have previous work experience. This shortcoming hampered an optimal appraisal of their work-related self-efficacy.

Lastly, in the follow-up of the control group we did not distinguish between sheltered paid employment and competitive paid employment. In the intervention group, we differentiated between these outcomes. Consequently, the outcome measure paid employment was less comparable between the two groups.

RECOMMENDATIONS FOR FUTURE RESEARCH

We made progress in researching the content and effectiveness of self-management support and vocational support. However, more research is needed to further optimize studies on these themes.

Part 1: Self-management support

Based on the first part of this thesis, we recommend to further develop or improve generic outcome measures that match with the content and characteristics of the self-management intervention. The International Consortium for Health Outcomes Measurement (ICHOM), for example, defined global Sets of Patient-Centered Outcome Measures that can be internationally used to measure health outcomes [55]. When all rehabilitation professionals would apply the same outcome measurements, comparisons of the results of different interventions on these outcomes would be more valid.

Regarding a subgroup of young people with chronic physical conditions who are vulnerable for a delay in psychosocial development, we recommended to specify the needs for professional support, so that psychosocial development interventions can be further tailored.

Part 2: Vocational support

Our study on the effectiveness of ‘At Work’ suggested several recommendations for future research. First, it is important to further optimize studies on the effectiveness of vocational rehabilitation interventions. The research design we used in our study, a controlled trial, may not be the most appropriate to this aim. A better option would be performing comparative effectiveness studies aimed at generating evidence from cohorts in real-life settings. The above-mentioned ICHOM Sets of Patient-Centered Outcome Measures can be used in comparative effectiveness studies. In addition, we recommended to develop a valid measurement instrument addressing a person’s preparedness of entering the competitive labor market, which so far is lacking.

Second, it is important to identify subgroups of young adults with chronic physical conditions with different needs for vocational support– from limited support to intensive programs. This would enable to further tailor vocational support and to redefine inclusion criteria for an intensive and multidisciplinary intervention such as the ‘At Work’ program. Well-defined inclusion criteria for vocational support will also help to improve effectiveness studies.

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Summary

Through advancements in healthcare and medication, more children with chronic physical conditions reach adulthood. Developing into an adult is very challenging for this subgroup. Adaptive tasks related to living with a chronic condition can hinder the accomplishment of age-appropriate developmental tasks. The psychosocial development of adolescents and young adults with chronic physical conditions lags behind compared to healthy age mates. It is important to educate them and to train skills for independent living with a chronic physical condition. Supporting young people with chronic physical conditions in their self-management is crucial. However, systematic insights into the content, effective intervention elements and effectiveness of self-management support are lacking. Therefore, part one of this thesis (Chapter 2-4) explored the content, effective intervention elements and effectiveness of self-management support for children, adolescents, and young adults with chronic physical conditions. In addition, we investigated the characteristics of a subgroup of adolescents with chronic physical conditions who are vulnerable for a delay in psychosocial development. It is important to identify this subgroup, in order to provide timely self-management support.

In the transition to adulthood, finding suitable work is challenging for young adults with chronic physical conditions. They reported several barriers for finding and maintaining employment: problems with transport to work, limited accessibility of buildings, limited adaption of workplaces or working conditions, discrimination and low self-esteem. The work participation of young adults with chronic physical conditions is low compared to healthy age-mates. Part two of this thesis (Chapter 5-8) addressed vocational rehabilitation interventions for young adults with chronic physical conditions. In this part we investigated the effectiveness of vocational support in general and of the intervention 'At Work' specifically.

Chapter 2 reported on a systematic literature review on the content and intervention elements of self-management support for young people (aged 7-25 yrs) with chronic physical conditions. Eighty-six studies were included. Self-management interventions are mostly focused on medical management (46.2%), for example self-monitoring of blood glucose values in diabetes, planning of doctor's appointments, and knowledge of medical aspects of the chronic condition. Hardly any attention was paid to emotion management (2.6%), such as self-confidence, developing a positive body image and stress management. Also, little attention was paid to role management (6.4%), such as in participating in sports, relationships, leisure activities, or communicating with others. Most of the included studies did not report on the theoretical base of the interventions (67.9%). The content of self-management interventions was not linked to specific intervention elements, such as setting (e.g. out-patient clinic, clinic or at home) or format (e.g. individual sessions, group sessions or a combination of both). Most

self-management interventions included education and/or skills training (43.2%) or telemonitoring (17.3%).

Chapter 3 described a systematic literature review on the effectiveness of self-management support and effective intervention elements. Forty-two randomized controlled were included. Because of the heterogeneity of intervention formats, and the heterogeneity of study outcomes and measurement instruments, the quality of evidence was low. Moderate evidence was found for the effectiveness of self-management interventions on disease knowledge and treatment adherence. Self-management interventions focused on symptom reduction and school absence were not effective. The literature review showed that interventions focused at dealing with the chronic conditions in daily life should preferably be delivered individually. For that purpose, an online format facilitating peer-support seemed to be appropriate and effective. For self-management interventions to improve treatment adherence, several intervention elements seemed to be appropriate and effective, such as a focus on medical management, an individual format, treatment in a clinical setting or at home provided by a monodisciplinary team.

Chapter 4 is focused on early predictors for claiming disability benefits by young adults with chronic physical conditions. Young adults who claimed disability benefits reported during adolescence more severe physical disabilities, followed less often special education, and indicated a higher absenteeism at school/work, a greater impact of the condition on school functioning, and a low health-related quality of life. In young adulthood, they reported a greater impact of their chronic condition on their school/work career and a lower quality of life as compared to young adults with chronic conditions who do not receive disability benefits.

Chapter 5 described a systematic literature review on the characteristics and effectiveness of vocational rehabilitation interventions aimed at improving work participation of people with chronic physical conditions. Thirty studies were included. Results showed that content and characteristics of vocational rehabilitation interventions varied widely. We appraised the effectiveness of vocational rehabilitation interventions on three outcomes of vocational participation. Results showed positive effects of vocational rehabilitation interventions on *work status* and *work attitude*, but the quality of evidence for these effects was low to very low. Vocational rehabilitation interventions were not effective on *work productivity*; in this respect the quality of evidence was low. Moderate evidence was found for the effectiveness of interventions that included multiple intervention components and were delivered individually, whether or not combined with group sessions.

Chapter 6 till 8 reports on the evaluation of the Dutch vocational rehabilitation intervention 'At Work'. This intervention is focused on young adults with chronic physical conditions who experience problems in finding and maintaining competitive employment after finishing post-secondary education. This program consist of six group-sessions followed by an individual coaching trajectory of one year. This intervention is delivered by an occupational therapist at an out-patient rehabilitation clinic and a job coach of a re-integration agency.

Chapter 6 evaluated the participant-perceived beneficial attributes of the 'At Work' program using semi-structured interviews (n=19) with former intervention participants. They mentioned peer-support and the job interview training as beneficial attributes, since these increased their self-esteem and self-efficacy. They indicated that their improved self-esteem and self-efficacy helped them to find and maintain employment. Former intervention participants mentioned that the support provided by the job coach, like the arrangement of adjusted working conditions and job placement assistance, helped them to find and maintain employment. Last, the education provided to the supervisor at work was mentioned as a beneficial attribute of the 'At Work' program, since it positively influenced the employer's or supervisor's attitude. Former intervention participants also recommended to include more communication skills training in the intervention. They argued that this training might help them to be more open and assertive at work, which will contribute to job retention.

Chapter 7 researched the effects of the 'At work' program that is aimed at improving the competitive employment of young adults with chronic physical conditions. Employment rates at baseline, post-interventions and two years follow up of a cohort of 'At Work' participants (n=90) were compared to national reference data of young adults with chronic physical conditions. At the start of the intervention, employment rates of 'At Work' participations were significant lower compared to national reference data (10.0% versus 52.9%). Employment rates of 'At Work' participants significantly increased to 42.2% at two years follow-up, and approached the employment rate of the reference cohort (52.9%).

Chapter 8 described a controlled clinical trial on the effectiveness of the 'At Work' program on outcome measures referring to work-readiness and work status, namely self-efficacy in solving work- and disease- related problems and general self-efficacy, and having paid employment. Participants in the intervention condition (n=49) were recruited at three outpatient rehabilitation clinics that delivered this specific intervention. Other outpatient rehabilitation clinics in the Netherlands recruited young adults for the control group (n=39). In the intervention group, scores on all outcome measures

increased over time, but this was also the case for the control group. Still, general self-efficacy tended to increase more over time in the intervention group as compared to the control group.

Chapter 9, the general discussion, presents the most important results of the studies in this thesis, discussed the results in relation to the current scientific literature, and described implications for future clinical practice.

Up to now, self-management interventions are mainly focused on medical management. It is important that transitional care also pays attention to emotional aspects and psychosocial consequences of living with a chronic physical condition. We concluded that healthcare teams for young adults should early monitor the psychosocial development of young people with chronic physical conditions, in order to timely start self-management interventions in case of delay in development. This is especially important for subgroups vulnerable for a delay in psychosocial development. With regard to vocational support, the 'At Work' program is a promising intervention for young adults with chronic physical conditions to achieve competitive employment after finishing their post-secondary education. Our evaluation studies pointed towards positive results of this intervention. Starting from a very disadvantaged position, more than 40% of the participants had paid work at two years follow-up. In a controlled trial, however, we did not find significant differences in treatment effects of 'At Work' as compared to usual care. Possible reasons for the lack of evidence are discussed. Former intervention participants valued the role playing activities, real world experimental opportunities and peer-support as important elements of the 'At Work' program, since these elements were beneficial to improve their self-efficacy and life skills. In order to foster the learning process in authentic learning situations it is important to provide job coaching immediately in the situation concerned. For this purpose, incorporating an online peer support format into the jobcoaching can be a useful addition.

SAMENVATTING

Door verbeterde zorg en medicatie bereiken steeds meer kinderen met chronisch lichamelijke aandoeningen de volwassenheid. Volwassen worden is een extra uitdaging voor deze groep. Het leven met een chronische aandoening kan soms interfereren met het behalen van ontwikkelingsmijlpalen. De psychosociale ontwikkeling van adolescenten en jongvolwassenen met chronisch lichamelijke aandoeningen loopt vaak achter in vergelijking met gezonde leeftijdsgenoten. Het is belangrijk hen vroegtijdig de kennis mee te geven en de vaardigheden te leren die nodig zijn voor het inpassen van de chronische aandoening in het dagelijks leven. Het is essentieel de adolescenten met chronisch lichamelijke aandoeningen te ondersteunen in hun zelfmanagement. Echter inzicht in de inhoud, in effectieve interventie elementen en effectiviteit van zelfmanagement ondersteuning ontbreekt. Deel 1 van dit proefschrift (hoofdstukken 2-4) verkent daarom deze aspecten van zelfmanagement support voor kinderen, adolescenten en jongvolwassenen met chronisch lichamelijke aandoeningen. Daarnaast onderzoekt deel 1 de kenmerken van een subgroep van adolescenten met chronisch lichamelijke aandoeningen die verhoogd risico heeft op vertraging in de psychosociale ontwikkeling. Het is belangrijk deze groep te herkennen en hen tijdig zelfmanagement ondersteuning aan te bieden.

Op de weg naar volwassenheid is het vinden van werk een grote uitdaging voor jongvolwassenen met chronisch lichamelijke aandoeningen. Zij noemen verschillende barrières voor het vinden en behouden van werk: problemen met vervoer naar werk, beperkte toegang tot gebouwen, beperkte aanpassing van fysieke werkomgeving of arbeidsvoorwaarden, discriminatie of laag zelfvertrouwen. De arbeidsparticipatie van jongvolwassenen met chronisch lichamelijke aandoeningen is dan ook lager dan bij gezonde leeftijdsgenoten. Deel 2 (hoofdstukken 5-8) richt zich op interventies om adolescenten en jongvolwassenen met chronisch lichamelijke aandoeningen naar werk te begeleiden. Deel 2 onderzoekt de effectiviteit van deze interventies op (re)integratie naar werk in het algemeen en van de interventie 'Aan het werk' in het bijzonder.

Hoofdstuk 2 gaat in op een systematische literatuurstudie naar de inhoud en interventie onderdelen van zelfmanagement ondersteuning voor kinderen en jongeren (7-25 jaar) met chronisch lichamelijke aandoeningen. Zesentachtig studies zijn geïncludeerd. De zelfmanagement interventies zijn meestal gericht op medisch management (46.2%), wat refereert naar taken of onderdelen zoals het zelf monitoren van bloedwaardes, het regelen van doktersafspraken, of kennis over de medische aspecten van de aandoening. Binnen de zelfmanagement interventies is er minder aandacht voor emotie management (2.6%), zoals zelfvertrouwen, het ontwikkelen van een positief zelfbeeld

en stressmanagement. Ook is er binnen de interventies weinig aandacht voor het managen van sociale rollen (6.4%), zoals in sport, vriendschappen, vrije tijd, of communicatie met anderen. De theoretische onderbouwing van de zelfmanagement interventies is vaak niet gerapporteerd (67.9%). De inhoud van de zelfmanagement interventies is niet gelinkt aan specifieke interventie onderdelen of kenmerken van de interventie, zoals setting (polikliniek, klinische setting, of thuis) of format (zoals individueel, online). De meeste zelfmanagement interventies zijn gericht op educatie en/of vaardigheidstraining (43.2%), of eHealth ondersteuning (17.3%).

Hoofdstuk 3 beschrijft een systematische literatuurstudie naar de effectiviteit van zelfmanagement support en effectieve interventie onderdelen. Tweeënveertig gerandomiseerde studies zijn geïnccludeerd. Door de heterogeniteit in interventies en de variatie in de gehanteerde uitkomstmaten en meetinstrumenten, is de kwaliteit van het bewijs voor effectiviteit laag. Er is matige evidence voor de effectiviteit van zelfmanagement interventies gericht op kennis over de ziekte en therapietrouw. Zelfmanagement interventies gericht op het verminderen van symptomen en school absentie blijken niet effectief te zijn. Het review laat zien dat interventies gericht op het omgaan met de chronische aandoening het beste individueel kunnen worden aangeboden. Een online format dat ondersteuning van lotgenoten faciliteert blijkt daarvoor passend en effectief. Voor zelfmanagement interventies gericht op het verbeteren van therapietrouw lijken focus op medisch management, een individueel format, een klinische setting of een thuis-interventie door een monodisciplinair team de meest passende en effectieve elementen te zijn.

Hoofdstuk 4 focust op vroege voorspellers voor het aanvragen van een Wajong uitkering door jongvolwassenen met chronisch lichamelijke aandoeningen. Jongvolwassenen die een Wajong uitkering aanvragen hadden tijdens de adolescentie vaker een ernstige fysieke beperking, volgden minder vaak speciaal onderwijs, verzuimden vaker van school of werk, en rapporteerden een lagere kwaliteit van leven. Als jongvolwassene rapporteren ze een grotere impact van de chronische aandoening op hun loopbaan en een lagere kwaliteit van leven dan jongvolwassenen met een chronische lichamelijke aandoening die geen Wajong-uitkering hebben.

Hoofdstuk 5 beschrijft een systematische literatuurstudie naar de kenmerken en effectiviteit van interventies gericht op het verbeteren van de arbeidsparticipatie van mensen met een chronische lichamelijke aandoening. Dertig studies zijn geïnccludeerd. De resultaten laten zien dat interventies om de arbeidsparticipatie te bevorderen sterk verschillen wat betreft inhoud en organisatie. We beoordeelden de effectiviteit op drie uitkomsten van arbeidsparticipatie. Voor het verbeteren van *werkstatus* en *werkhouding*

lijken de interventies effectief te zijn, al is de kwaliteit van dit bewijs zeer laag. Interventies gericht op het verbeteren van *arbeidsproductiviteit* blijken niet effectief; de kwaliteit van dit bewijs is laag. We vonden matig bewijs voor de effectiviteit van interventies die uit meerdere componenten bestaan en die individueel worden aangeboden, of in combinatie van individuele begeleiding en in groepsverband.

In **hoofdstuk 6 tot en met 8** wordt de Nederlandse interventie ‘Aan het werk’ geëvalueerd. Deze interventie richt zich op jongvolwassenen met chronisch lichamelijke aandoeningen (16-27 jaar) die na het afronden van hun opleiding problemen ervaren met het vinden van werk op de reguliere arbeidsmarkt. De interventie start met zes groepssessies gevolgd door individuele coaching gedurende een jaar. Het programma wordt aangeboden door een ergotherapeut werkzaam in de revalidatie en een jobcoach van een re-integratie bedrijf.

Hoofdstuk 6 evalueert door middel van semigestructureerde interviews met oud-deelnemers (n=19) de door hen ervaren effecten en werkzame onderdelen van de interventie ‘Aan het werk’. Zij geven aan dat lotgenotencontact en sollicitatietraining werkzame en passende onderdelen zijn van het programma, die volgens hen bijdragen aan meer zelfvertrouwen en geloof in eigen kunnen. Het verbeterde zelfvertrouwen heeft hen geholpen bij het vinden en behouden van werk. De geboden ondersteuning door de jobcoach bij het regelen van aanpassingen in arbeidsomstandigheden en arbeidsvoorwaarden, en bij het vinden van passend werk, heeft volgens de oud-deelnemers bijgedragen aan het daadwerkelijk vinden en behouden van betaald werk. Ze benoemen daarom deze interventie onderdelen ook als ervaren werkzaam. Tenslotte ervaren ze de educatie van de jobcoach aan de leidinggevende op het werk over de chronische aandoening en de gevolgen daarvan voor het werk als een werkzaam onderdeel van de interventie. Ze geven aan dat deze educatie de vaak negatieve attitude van een werkgever positief beïnvloedt. De oud-deelnemers hebben ook bepaalde ondersteuning gemist in het programma ‘Aan het werk’, zoals meer aandacht en het daadwerkelijk trainen van hun communicatieve vaardigheden. Deze training zou hen meer open over de aandoening en assertief hebben gemaakt, wat volgens de oud-deelnemers bij had kunnen dragen aan het behouden van hun baan.

Hoofdstuk 7 onderzoekt de effecten van de interventie ‘Aan het werk’ gericht op de arbeidsparticipatie van jongvolwassenen met chronisch lichamelijke aandoeningen. De arbeidsparticipatie bij start van de interventie, na afloop en op 2 jaar follow-up van een cohort deelnemers aan de interventie ‘Aan het werk’ (n=90) is vergeleken met de arbeidsparticipatie van Nederlandse referentie data van jongvolwassenen met chronisch lichamelijke aandoeningen van het CBS. Bij start van de interventie ‘Aan het werk’

hebben de deelnemers minder vaak betaald werk dan jongvolwassenen met chronisch lichamelijke aandoeningen uit het referentie cohort (10.0% versus 52.9%). Het percentage deelnemers aan de interventie dat betaald werk heeft neemt significant toe over twee jaar tijd naar 42.2% en benadert dat van het referentie cohort (52.9%).

Hoofdstuk 8 beschrijft een gecontroleerde interventie studie naar de effecten van de interventie ‘Aan het werk’ op uitkomsten met betrekking tot arbeidsgereedheid en werkstatus, namelijk geloof in eigen kunnen (voor het oplossen van werk- en aandoening gerelateerde problemen of in algemene zin) en het hebben van betaald werk. Deelnemers voor de interventie groep (n=49) zijn geworven op de drie poliklinieken revalidatiegeneeskunde die de interventie ‘Aan het werk’ aanbieden. Andere poliklinieken revalidatiegeneeskunde die deze interventie niet aanbieden hebben jongvolwassenen benaderd voor de controle groep (n=39). De interventiegroep verbetert op alle uitkomstmaten over de tijd, maar dit gebeurde ook in de controle groep. Wel lijkt in de interventiegroep het geloof in eigen kunnen in het algemeen sterker te verbeteren over de tijd, vergeleken met de controle groep.

Hoofdstuk 9 – de algemene discussie – bespreekt de belangrijkste resultaten van het gehele onderzoek, bediscussieert deze resultaten in relatie tot de huidige ontwikkelingen en de literatuur, en beschrijft implicaties voor de praktijk.

Zelfmanagement interventies zijn tot nu toe voornamelijk gericht op medisch management. Het is belangrijk dat transitiezorg zich daarnaast ook richt op de emotionele aspecten en psychosociale gevolgen van het leven met een chronische aandoening. De conclusie is dat zorgteams voor adolescenten en jongvolwassen de psychosociale ontwikkeling vroegtijdig moeten monitoren, zodat bij vertraging in de ontwikkeling tijdig passende interventies kunnen worden aangeboden. Dit is zeker van belang bij subgroepen van adolescenten die een verhoogde kans hebben op een vertraging in de psychosociale ontwikkeling. De interventie ‘Aan het werk’ is een veelbelovend programma om jongvolwassenen met chronisch lichamelijke aandoeningen naar regulier werk te begeleiden na afronding van de opleiding. De door ons uitgevoerde evaluatiestudies wijzen op positieve resultaten van de interventie. Startend vanuit een zeer lage arbeidsparticipatie, heeft ruim 40 procent van de deelnemers na 2 jaar betaald werk. In een gecontroleerde klinische studie hebben we echter geen hard bewijs gevonden voor de effectiviteit van de interventie vergeleken met gebruikelijke zorg. Mogelijke redenen hiervoor worden besproken. Oud-deelnemers waarderen de rollenspellen (zoals sollicitatietraining), gecreëerde authentieke leersituaties en het lotgenoten contact als waardevolle onderdelen van het programma, omdat deze bijdragen aan het zelfvertrouwen voor werk en de ontwikkeling van ‘life skills’. Om het leerproces in werksituaties

te bevorderen is het belangrijk dat de jobcoach direct kan interveniëren, waarvoor het gebruik van 'e-mentoring' een bruikbare toevoeging kan zijn.



Appendices

DANKWOORD

Nu het einde van het promotietraject nadert, kijk ik terug op de afgelopen jaren. Het traject heb ik ervaren als een leerzaam proces, waarbij ik uitgedaagd ben op inhoud, en mij op zowel persoonlijk als professioneel vlak heb mogen ontwikkelen. Dankbaar ben ik voor de kansen die ik heb gekregen en de manier waarop dit hele proces mij heeft gevormd. Ik heb dit proefschrift niet kunnen afronden zonder de hulp en steun van alle mensen om mij heen. Hartelijk dank daarvoor! Een aantal mensen wil ik in het in het bijzonder bedanken.

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Marjolijn

PHD PORTFOLIO

Name PhD student: Marjolijn Irene Bal
Erasmus MC Department: Rehabilitation Medicine

PhD period: 2012-2022
Promotor: Dr. Marij Roebroek
Supervisor: Dr. Harald Miedema

1. PhD training

	Year	Workload (Hours)
Research skills and in-depth courses		
• Systematic review Workshop KU Leuven	2012	24
• Beyond Simple Randomization Erasmus MC	2012	8
• Atlas.ti for qualitative data analysis Evers Research & Training	2012	16
• Effective methods to change behavior Maastricht University	2013	4
• Biomedical English writing course Molecular Medicine Postgraduate School	2014	56
• Multilevel statistical analyses Rotterdam University of Applied Sciences	2014	16
• Scientific Integrity Erasmus MC	2014	8
• Focus group discussions Evers Research & Training	2015	16
Symposia		
• Transition of young people with chronic conditions from special secondary education to vocational education Reade Amsterdam	2012	4
• Symposium Quality and implementation Erasmus MC	2013	4
• Symposia Lifespan care and Transition Rijndam Rehabilitation & Erasmus MC	2013 & 2014	4
• Symposium Kracht van Kennis van Arbeidsdeskundigen Kenniscentrum (AKC), Amsterdam, Nederland	2014	8
Conferences		
• National Program Zelfmanagement congress Utrecht, Netherlands	2012	8
• Cohere congress Groningen, the Netherlands	2014	16
• 18e IAAH congress (International Association Adolescent Health) Parijs, France	2014	24
• Dutch Congress of Rehabilitation Medicine (DCRM) Rotterdam, Nederland	2014	8
• Work 2015 congress Turku, Finland	2015	24

Research groups

- | | | |
|---|-------------|----|
| • Research meetings Expertise Group Self-management and Participation, Rotterdam University of Applied Sciences | 2012- 2018 | 50 |
| • Research meetings department of Rehabilitation Medicine, Erasmus MC | 2012 - 2018 | 90 |

Oral presentationsA) Presentations at Conferences*International*

- | | | |
|--|------|---|
| • Experiences with a multidisciplinary group intervention to improve work participation among adolescents and young adults with physical disabilities; 18e IAAH Congress (International Association Adolescent Health), Parijs, France | 2014 | 8 |
| • Improving educational and vocational participation among young adults with chronic physical conditions; Work 2015, Turku, Finland | 2015 | 8 |

National

- | | | |
|---|------|---|
| • Experiences with a multidisciplinary group intervention to improve work participation among young adults with physical disabilities; Cohere congress, Groningen, the Netherlands | 2014 | 8 |
| • Professionele ondersteuning bij school- en arbeidsparticipatie van jongvolwassenen met een chronisch lichamelijke aandoening; Symposium Kracht van Kennis van Arbeidsdeskundigen Kenniscentrum (AKC), Amsterdam, Nederland | 2015 | 4 |
| • Outcomes of a multidisciplinary group intervention to improve work participation among young adults with physical disabilities: A mixed methods study; Dutch Congress of Rehabilitation Medicine (DCRM), Rotterdam, Nederland | 2014 | 8 |
| • Uitkomsten van de Interventie TraJect aan het werk?!. Presentatie Symposium TraJect aan het werk?!. Erasmus MC, Rotterdam, Nederlands | 2015 | 4 |

B) Presentations at Research Meetings

- | | | |
|--|------------------------|----|
| • Effectiviteitsstudie module aan het werk | 2012, 2014, 2018, 2021 | 22 |
| • Studie naar de ervaren werkzame onderdelen module aan het werk | 2013 | 4 |
| • Studie naar ervaringen met de (voorbereiding op de) transitie van speciaal onderwijs naar beroepsonderwijs van adolescenten met een chronische lichamelijke aandoening | 2015, 2018 | 8 |
| • Studie naar de psychosociale ontwikkeling van jongeren met chronische aandoeningen: Vergelijking tussen jongeren met en zonder Wajong uitkering | 2020 | 4 |

C) Presentations for consortiumpartners

- | | | |
|---|------|---|
| • TransitieNet expert meeting, Rotterdam | 2012 | 4 |
| • Reade Wetenschappelijk Overleg revalidatieartsen, Amsterdam | 2012 | 4 |
| • Studiedag Kinderrevalidatie Rijndam Revalidatie Rotterdam | 2014 | 8 |
| • Studiedag Rijndam/mytyschool de Brug, Rotterdam | 2015 | 4 |
| • Studiedag Mytyschool de Brug, Rotterdam | 2015 | 4 |
| • Transitieavond 'World Cafe' Mytyschool de Brug, Rotterdam | 2015 | 4 |

Other

- | | | |
|-----------------------------------|----------|----|
| • Reviewer of scientific journals | 2016-now | 20 |
|-----------------------------------|----------|----|

2. Teaching

Training

- | | | |
|---|-----------|-----|
| • Didactics
Rotterdam University of Applied Sciences | 2012-2013 | 300 |
|---|-----------|-----|

Lecturing

- | | | |
|---|------------|-----|
| • Bachelor Instituut voor Gezondheidszorg, Hogeschool Rotterdam:
diverse bijdragen aan minoronderwijs en projectbegeleiding (minor
Kind & Jeugd en Arbeid, Gezondheid en welzijn) | 2012-2013 | 16 |
| • Bachelor Ergotherapie, Hogeschool Rotterdam: | | |
| - Wetenschappelijk redeneren | 2012 – now | 360 |
| - Onderzoeksvaardigheden | 2012 – now | 460 |

Supervising trainees

- | | | |
|---|-----------|----|
| • Supervising interns and trainees Bachelor and Master Theses | 2012-2018 | 72 |
|---|-----------|----|

Total (Hours/ ECTS)	1677 hours/ 60 ECTS
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LIST OF PUBLICATIONS (NOT INCLUDED IN THIS THESIS)

International

- Saan M.C., Boeije H.R., Sattoe J.N.T., Bal M.I., Missler M., van Wesel F. (2015) Recording and accounting for stakeholder involvement in systematic reviews. *Health Information and Libraries Journal*, 32(2): 95-106.

National

- Bal MI, Hilberink SR, Verhoef JAC, Miedema HS, Roebroek ME. (2012). *Eindverslag proefimplementatie TraJect aan het werk*. Rotterdam: Erasmus MC/ Kenniscentrum Zorginnovatie
- Van Hagendijk MM, van Rossum NA, Miedema HS, Bal MI. (2015). Van participatiewet naar duurzame arbeidsparticipatie door mensen met een chronische aandoening: een kwalitatieve studie naar behoeften van werkgevers aan ondersteuning bij de uitvoer van de participatiewet. *Sociaal bestek*, 1, 46-47
- Van Staa AL, Miedema H, Sattoe J, Bal M, Roelofs P, Hilberink S, Ista E, Roebroek M, Peeters M. (2015). *SPIL eindrapportage*. Rotterdam: Kenniscentrum zorginnovatie
- Verhoef JAC, Miedema HS, Bal MI, Roebroek ME. (2021). Interventies om de arbeidsparticipatie van mensen met een chronische lichamelijke aandoening te bevorderen. *Nederlands tijdschrift voor revalidatiegeneeskunde*, 42(1), 12-16.
- Bul K, Bal M, Kuiper C, & Vuijk P. (2021). Naar de middelbare school als je ass hebt. overstap vergt veel steun en begeleiding. *Kind en Adolescent Praktijk*, 20(3), 6-14.

CURRICULUM VITAE

Marjolijn Bal was born in Maassluis on the 23rd of January 1989. She followed her secondary education at Lentiz Revislyceum (2001-2007). Marjolijn studied Psychology and got her Bachelor's degree in 2010. In 2011, she graduated in Social and Organizational Psychology at Leiden University. In 2011, she started working as a researcher at Research Centre Innovations in Care, Rotterdam University of Applied Sciences. After 1 year working as a researcher, in 2012, she started her PhD project at Erasmus University Medical Department of Rehabilitation Medicine. Marjolijn's research focuses on work participation of young adults with chronic physical conditions. Currently, she is working as a lecturer at the Rotterdam University of Applied Sciences, School of Health Care Studies (occupational therapy) and as a researcher at Research Centre Innovations in Care.

