Upper limb absence
Postema, Sietke

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Document Version
Publisher's PDF, also known as Version of record

Publication date:
2017

Link to publication in University of Groningen/UMCG research database

Citation for published version (APA):

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Chapter 8

General discussion
The principle aim of this thesis was to explore the effects of upper limb absence (ULA), due to a congenital reduction deficiency (RD) or an acquired amputation (AA), on the remaining body functions and structures, on prevalence and characteristics of musculoskeletal complaints (MSC), and functional capacity. In chapter 2 it was shown that remaining body structures and functions of children with RD are affected by ULA, which remain, and sometimes become more evident, in adulthood. Asymmetry of the trunk and upper limbs were found, as well as restrictions in range of motion of the affected limb. Overall disability was greater in individuals with RD; however work-related disability did not differ between individuals with RD and matched controls. A relatively low rate of MSC (30%) was found. Assessment of prevalence and characteristics of MSC were described in chapter 3; a significant higher prevalence of MSC in individuals with ULA (57% point prevalence) compared to controls, and no difference in prevalence of MSC between individuals with RD and AA was found. Clinically relevant predictors for MSC were middle age, being divorced or widowed, and lower mental health. Disability was predicted by higher age, more pain, lower general and mental health, and not using a prosthesis. In chapter 4, presence of MSC was not related to work participation, but MSC-related pain was the most important predictor for decreased work productivity. Male sex, younger age, a medium or high level of education, prosthesis use, and good general health were predictors for work participation. Individuals with RD were more often employed compared to individuals with AA. However, when employed, work productivity did not differ between these two groups. The development and pilot testing of the functional capacity evaluation – one-handed (FCE-OH) is reported in chapter 5. This instrument examines the functional capacity of the upper limbs of one-handed individuals. Results of pilot testing demonstrated generally similar functional capacity for individuals with ULA and matched controls, and it was hypothesized that this, combined with a higher physical load on the unaffected limb, might actually reflect a relative deficit of functional capacity in one-handed individuals. Test-retest reliability of this instrument was acceptable in five out of eight items (chapter 6). Furthermore, this instrument was proven to be safe in use. Finally, the development of a qualitative scoring system for rating compensatory shoulder and trunk movements in upper limb prosthesis users during the performance of the FCE-OH was described in chapter 7; the scale has sufficient intra- and interrater reliability. Feasibility of the scoring system was established.

This final chapter provides a perspective on the main findings of the previous chapters, describes the (clinical) implications of the studies conducted, and gives suggestions regarding further research.

ULA is more than the missing of a limb

This thesis shows that an RD of an upper limb affects the remaining body structures and functions. Structural asymmetries of body halves, and restrictions in range of joint
motion of individuals with ULA may have arisen as a consequence of the insult that caused RD, or due to under- and over-use of particular muscles (chapter 2). Body functions and structures of individuals with AA were not examined in this thesis, but from previous research it is known that reduced shoulder range of motion occurs in individuals with AA as well.16

Another effect of ULA is the increased risk on development of MSC. Several studies across the world showed that individuals with ULA are prone to MSC,4–10 and in chapter 3 it was established that Dutch individuals with ULA are no exception. In individuals with ULA, MSC were frequently present at multiple sites and often lasted for more than a year. Furthermore, pain intensity and disability were higher in individuals with ULA compared to two-handed individuals (chapter 3); emphasizing the severity of the problem. No difference in prevalence of MSC between individuals with RD and AA was found (chapter 3), which is in line with the results of Burger and Vidmar.10 Apparently, despite the natural development of compensation strategies134 and the few limitations children and youngsters with RD experience,134,135 they remain prone to MSC in later life.

Personal and environmental factors predicting MSC

Various models regarding the pathomechanisms of MSC are available, and many of these describe a multifactorial original, presuming an interactive effect between (work-related) psychosocial and biomechanical strain.108 Results of this thesis suggest that also in individuals with ULA both psychosocial and biomechanical risk factors may play a role in the development of MSC.

Psychosocial risk factors

Psychosocial factors, such as depression, pain catastrophizing cognitions, coping, and perceived social support can predict physical and psychological functioning.136–138 Although coping styles were no predictors for presence of MSC and MSC-related disability, mental health was associated with both (chapter 3). The causal relationship between mental health and MSC has not been established in this research, but it is imaginable that presence of pain negatively influences mental well-being. For example, pain seems to be related to lower health related quality of life in individuals with ULA.7 Moreover, in a study among individuals with upper and lower limb amputations, depressive symptoms were found to be a significant predictor of level of pain intensity and bothersomeness.6 However, decreased mental health may also predispose to (awareness of) MSC. For example, depression was reported as the largest hazard ratio for newly developed low back pain in a 3-year longitudinal study.139 Bongers et al described several explanations on how perceived (work) stress could induce or exacerbate MSC.140 Firstly, it might be that psychosocial factors directly influence the mechanical load through changes in posture, movement and exerted forces. Moreover, stress may increase the muscle tension, and may enhance the perception of symptoms or reduce the capacity to cope with these symptoms. Both stress and pain are modulated in the same part of the brain, the amygdale, which is involved in assigning emotional significance to environmental
information and triggering adapted physiological, behavioral and affective responses. The amygdale also contributes to pain processes, and persistent and chronic pain may alter amygdale activity. Although the exact pathways remain unclear, results of studies focusing on the amygdale suggest a strong relation between pain and emotion. It is therefore not a coincidence that a psychologist or psychomotor therapist is often part of the contemporary multidisciplinary rehabilitation team for chronic pain patients.

Furthermore, in individuals with ULA, behavior may play a role in development of MSC as well, as half of the individuals with MSC stated that presence of MSC is obvious when having ULA (chapter 3). This could lead to continuation of behavior that led to the development of MSC, and may prevent them from changing their behaviors or consulting a healthcare professional.

**Biomechanical risk factors** In this thesis, increased physical demands and compensatory movements are assumed to be related to the higher risk of MSC. This assumption arises from the epidemiologic evidence that repetition, posture, and produced force play a role in the development of MSC. Furthermore, it is known that when daily tasks are performed with a prosthetic limb, larger range of motions of torso, shoulder and elbow are necessary compared to tasks performed by an unaffected limb, and movements with a prosthesis are less fluent. When one grasps with a prosthesis decoupling of reaching and grasping occurs. Thus, using a prosthesis demands increased time, range of motion of torso, shoulder and elbow, energy expenditure, and cognitive load to plan alternative reaching strategies, and is related to a less efficient reach path and grasp pattern. Due to these demands from the affected side, activities will be executed by the unaffected limb whenever possible, thus increasing the physical load on the unaffected upper limb.

Higher physical loads and compensatory movements may result in altered patterns of muscle use, leading to muscle imbalance due to under- and over-use of particular muscles. Muscle imbalance may in turn lead to a self-perpetuating condition of weakening of certain muscles due to lack of use, while other muscles are overused and become hypertrophic, resulting in chronic pressure on nerves and reduced peripheral circulation. This may explain why individuals with ULA experience more often tingling compared to controls, when MSC is present (chapter 3). Higher physical loads may comprise increased repetition and force, which interact in a logarithmic manner; when low levels of force are required, many repetitions can be made until failure of tissue occurs. In this case, failure of tissue denotes the occurrence of precursors of MSC, such as microtrauma’s and inflammation. However, due to the logarithmic interaction, small increases in force requirements, decrease the number of repetitions until microtrauma’s and inflammation occur exponentially. Thus, this interaction effect of force and repetition has and additive effect on the risk of development of MSC.

In chapter 3 and 4 repetition, posture and force requirements of individuals with ULA were assessed with the upper extremity work demands scale. This scale, consisting of 7
questions, assesses the frequency of exposure to these determinants for MSC,\textsuperscript{76} and was significantly related to presence of MSC (chapter 3). This is a self-rated scale, and validation of this scale is necessary before well-founded conclusions can be drawn regarding the relationship between risk on MSC and upper extremity work demands.

**Consequences of MSC for disability**

Individuals with ULA have a high risk on MSC, which in turn may influence self-reported disability (chapter 3) and work productivity (chapter 4). Furthermore, presence of MSC is related to lower perceived general and mental health. Such a decrease of perceived general and mental health, as well as a higher pain score, is associated with higher self-reported disability (chapter 3).

Not surprisingly, individuals with ULA showed higher general disability scores on the disability of the arm, hand, and shoulder (DASH) questionnaire, which asks about symptoms and ability to perform tasks with the upper limb, compared to two-handed individuals (chapter 2). When MSC are present, individuals with ULA report higher levels of disability compared to able-bodied controls (chapter 3). This can be described as “dual disability”,\textsuperscript{28} in which disability caused by pain adds to the disability caused by one-handedness. In a study among over a hundred individuals with ULA, pain in the unaffected limb was related to a higher number of disability days (days individuals were kept form usual activities due to pain) compared to phantom limb pain, residual limb pain, and pain in the neck or back.\textsuperscript{7} This could be explained by the important role of the unaffected limb in daily functioning. In chapter 3, older age was related to increased “dual disability”, which is in line with previous findings that older individuals with ULA were more likely, compared to younger individuals, to be extremely bothered by pain in the unaffected limb.\textsuperscript{7}

**Consequences of MSC for work productivity**

Even though general disability was higher in individuals with RD compared to matched able-bodied controls (chapter 2), results on the work module of the DASH did not differ between both groups, suggesting that individuals with RD chose their work based on work abilities. In the Netherlands (chapter 4) and in Sweden,\textsuperscript{82} the rate of work participation of individuals with RD is similar to the general population. Depending on age at time of amputation, individuals with AA might not have had the opportunity to develop their career path based on their changed physical abilities, which may explain why changes in job and job tasks can be necessary after return to work (chapter 4).\textsuperscript{21} Nevertheless, when employed, individuals with RD and AA report equal work productivity, which did not differ from the work productivity of two-handed individuals (chapter 4). Work productivity was independent of physical work demands, which was confirmed in a study among Swedish adults with RD.\textsuperscript{82} Several youngsters performed mentally demanding work, while none of them rated their ability to work as being poor in relation to the physical, psychological and mental demands of their work. Presence of MSC was not related to work participation (chapter 4); however, MSC-related pain was the most important statistical
predictor of work productivity. Pain as an interference of work productivity was described previously in different patient groups with chronic musculoskeletal disorders.\textsuperscript{24-27} Health variables, particularly mental health, account for the largest proportion of explained variance in both work absence and performance. Moreover, several studies have found an association between severity of depression and productivity loss.\textsuperscript{147-150} Thus, in individuals with ULA, decreased work productivity can be directly caused by MSC-related pain, but may also be mediated by decreased mental health related to presence of MSC.

In conclusion, presence of MSC is associated with decreased health perception and increased disability. Individuals with RD may choose a career path and job based on their work abilities, and can be as productive as their two-handed colleagues. Even though individuals with AA might require change of work or work tasks, they too can be as productive as two-handed individuals. Physically demanding work is not avoided, and is not related to decreased work productivity. However, work productivity is negatively influenced by MSC-related pain.

Assessment of functional capacity

A functional capacity evaluation (FCE) can be defined as an evaluation of capacity of activities that is used to make recommendations for participation in work while considering the person’s body functions and structures, environmental factors, personal factors and health status.\textsuperscript{30} This thesis describes the development and reliability testing of an FCE for one-handed individuals (FCE-OH), which can be used to assess work capacity, give individuals with ULA appropriate advice regarding work participation, and measure outcomes of rehabilitation treatment. The FCE-OH consists of several tests, assessing overhead lifting, endurance of overhead working, repetitive reaching, fingertip dexterity, and handgrip strength. As individuals with ULA typically make compensatory movements in daily tasks, which are believed to play a role in the development of MSC, and currently no measurement tool exists to quantify these compensatory movements, a qualitative observational scale was developed (chapter 7). Once professionals are trained in the use of the instrument, this scale can be used to assess compensatory movements in individuals with ULA. It is hoped that assessment of compensatory movements may aid patients and professionals in determining optimal movement patterns, which are task-specific and depend on the motor characteristics of the patient. In addition, awareness of compensatory strategies among individuals with ULA is needed, in order to establish optimal movement patterns in daily life.

In chapter 5 participants from the Netherlands pilot tested the FCE-OH; while the participants, who were included in the repeatability study (chapter 6), were from Italy. Approximately 90\% of the Italian participants had transradial ULA. When comparing the Dutch participants with transradial ULA and the Italian participants, similar results were found for most FCE-OH tests. The repetitive reaching test was performed better by the Italians, while the fingertip dexterity test was performed slightly better by the Dutch. Overhead working endurance of individuals with ULA is similar to the results of patients
with back pain, whiplash associated disorders, and able-bodied adults. Overhead lifting capacity is lowest in individuals with ULA (8-11kg, chapter 5 and 6), ranging to almost 20kg in young able-bodied adults. Interestingly, individuals with ULA lifted close to similar weights whether or not a prosthesis was used. Although a prosthesis may increase functionality in daily life, through the ability of grasping and fixating of objects, it does not seem to enable individuals with ULA to increase lifting capacity. Bad grip with the prosthesis was the most mentioned reason not to lift more weight.

Functional capacity was generally similar between individuals with ULA and two-handed matched controls (chapter 5). It was hypothesized that this may actually denote a relative deficit of capacity, as one-handedness increases physical demands. According to the dose-response model, repeated and prolonged exertions require adaption of tissues to increase capacity. When tissues fail to adapt, dose tolerance decreases. An increased activity level of the tissues, without adequate adaptation, may explain why no difference was found between individuals with and without MSC (chapter 5).

The FCE-OH is safe in use, and most tests of the FCE-OH are reliable (chapter 6). For three tests further research is necessary in order to establish reliability. Professionals should be aware of learning effects and large limits of agreement; only changes outside the limits of agreement should be considered as real change. A training regarding the administration of the compensatory movement scale is advised (chapter 7). The current scale is suitable for therapists who are not experienced in assessment of compensatory movements; improvement of compensatory movement recognition may in time allow a more extended version of the current scale. The FCE-OH, including the qualitative observational scale for compensatory movements, could be a valuable instrument to assess functional capacity after injury and over time, give advice regarding appropriate work, and assess risk on and consequences of (sub)acute and chronic MSC. Furthermore, behavior during performance of an FCE-OH protocol may give important information about the patient’s efficacy and coping style.

Recommendations for treatment of individuals with ULA

The outcomes of this thesis yield several recommendations for treatment of individuals with ULA. First, this thesis shows that ULA is more than the missing of a limb, and attention for the remaining body functions and structures is thus warranted in rehabilitation treatment of individuals with ULA. The high risk on MSC, combined with an often chronic duration of complaints and increased disability, endorse the need for a preventive program. Individuals with ULA should be aware of the risk on MSC caused by one-handedness, they should have insight in possible compensatory strategies and the balance between physical capacity and physical demand, and be advised to alter their behaviors or consult a healthcare professional when complaints arise. Perhaps, a prevention program for MSC is necessary, including both consultations with rehabilitation professionals when called for, as well as self-management tools, aiming for awareness, an appropriate balance between physical capacity and work demands, and optimization of
compensatory strategies. Furthermore, resistance training of the neck, shoulder, and arm musculature might be beneficial for prevention and treatment of MSC, as concluded by a systematic review on the effectiveness of workplace interventions in the prevention of upper extremity MSC. Different theories on pathomechanisms of MSC support the hypothesis that physical training can improve muscle balance and capacity. In order to be effective and safe, this training should have the right balance between force, repetition, and rest.

Second, special attention is warranted for the older individual, as older age was associated with a higher risk on MSC, and increased MSC-related disability (chapter 3). Possibly, assessment of work capacity and work demands should be reevaluated in the aging individual. Collaboration between the rehabilitation and occupational medicine is warranted.

Last, as discussed, poor mental health could influence development of MSC in several ways, and depressive symptoms are a predictor for the level of pain and bothersomeness of the pain. Therefore, mental health should be monitored, and treatment should be offered when poor mental health is suspected.

Methodological considerations: strengths and limitations

The performed research provides new insights into the consequences of one-handedness on remaining body structures and functions. As far as I know, never before were results of physical examination of individuals with RD with a follow up of more than two decades presented. Furthermore, the study on MSC in individuals with ULA is the largest performed so far, executed on national level, and the only one including a significant number of individuals with RD, allowing for comparison of individuals with RD and AA. In addition, not only presence of MSC, but also consequences for disability and work participation and productivity were described. Moreover, this thesis described the development and reliability testing of the first measurement tool for functional capacity and compensatory movements of individuals with ULA. Studies were performed in three different countries (Sweden, the Netherlands, and Italy), and all but the reliability studies, included a control group.

Nevertheless, the performed studies also have limitations. The studies presented in chapter 4, 5 and 7 encompass a subgroup of the participants of the study presented in chapter 3. Therefore, some bias in representation of the total population of individuals with ULA may have influenced the results of multiple studies. Upper extremity work demands were assessed with a questionnaire, which is not the gold standard for assessment of physical work demands; neither has the scale been validated by comparing it with a gold standard. Thus, no solid conclusion regarding the relationship between work demands and risk on MSC can be drawn. Furthermore, in multiple studies heterogeneity of prosthesis use of individuals with ULA, due to changes in wearing time and type of prosthesis over time, made it unfeasible to make more precise evaluations of the effects of prosthesis use on body structures and functions, and on MSC. Moreover, the study in which the development of the
FCE-OH is described included 20 individuals with ULA. Based on the results of this study it was hypothesized that individuals with ULA have a relative deficit of capacity. The number of participants was sufficient for pilot testing of the FCE-OH, but does not allow conclusions regarding functional capacity of individuals with ULA. Further study limitations specific to each study are described in the corresponding chapters.

Recommendations for future research

The performed studies give several recommendations for further research. Future studies should aim to:

> Assess the validity of the upper extremity work demand scale by comparing the self-assessed questionnaire with a gold standard (such as observation or motion capture system). After validation of the upper extremity work demand scale, a study is warranted where individuals with ULA are matched to a two-handed colleague with a similar type of work, while upper extremity work demands of both are assessed. This may give insight in the added physical demands caused by one-handedness, allowing well-founded formulation of work recommendations.

> Validate the FCE-OH before use in rehabilitation care. Moreover, the reliability of three tests should be assessed in a larger population (repetitive reaching test, and fingertip dexterity test), or with a larger interval between measurements (overhead working test). Furthermore, the number of repetitions until a plateau is reached, and the amount of time to wear off learning effects of the repetitive reaching test and fingertip dexterity test should be determined.

> Study the hypothesis of relative deficit of functional capacity. This hypothesis was described in chapter 5, but the number of participants in this study did not allow conclusions regarding functional capacity of individuals with ULA.

> Improve degrees of freedom of prostheses. Compensatory movements are typically attributed to the limited degrees of freedom of prostheses. An increase of degrees of freedom may diminish compensatory movements. Moreover, feedback and improved fitting through osseointegration deserves further research, as it may improve the patients’ reliance on the prosthesis. Thus far, compensatory movements have been assessed in laboratory settings only. In order to establish the magnitude of compensatory movements, assessment of the influence of ULA on movement patterns in daily life, for example during work related tasks, is warranted.
Establish the causal relationship between MSC and MSC-predictors, such as mental health, upper extremity work demands, and prosthesis use in a longitudinal study. Results of such a study may give important information about necessary components of a (self-management) prevention program for MSC.

Final conclusions

Absence of an upper limb is more than just “missing a limb”; I have shown in this thesis that ULA also affects the remaining body structures and functions, is related to a higher risk on MSC, and influences functioning. Presence of MSC is a serious complication for individuals with ULA. MSC do not only occur more often in individuals with ULA compared to able-bodied individuals, but these individuals also report more pain, often have complaints on multiple bodily locations, and the duration of complaints is often long. Furthermore, presence of MSC negatively influences disability and work productivity. Both psychosocial and mechanical factors may play a role in the development of MSC. Cause of ULA, being RD or AA, was not a predictor for MSC or work participation. With the exception of the overhead lifting capacity, functional capacity of individuals with ULA seems to be equal to two-handed individuals. However, it is hypothesized that this may denote a relative deficit of capacity, as the unaffected limb of individuals with ULA may have to deal with increased physical demands. The FCE-OH and its compensatory movement scale may become valuable additions to patient assessment for execution of work related tasks by one-handed individuals.