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RESEARCH ARTICLE

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Musculoskeletal complaints and disability in a group of young adults with major congenital upper limb differences in The Netherlands

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ABSTRACT

Purpose: To determine prevalence of musculoskeletal complaints (MSCs) in adults with major congenital upper limb differences (CoULD) compared to able-bodied controls, and to examine associations of MSCs and disability with various biopsychosocial factors.

Materials and methods: Questionnaire-based cross-sectional study assessing MSCs, disability (using the Disabilities of the Arm, Shoulder and Hand questionnaire (DASH)), general and mental health status, physical work demands, and upper extremity range of motion.

Results: Seventy-one individuals with CoULD (participation rate: 41%) and 71 controls matched on age, gender, and education were included (49% female, mean age 28.9 years). Year prevalence of MSCs was significantly higher in the CoULD group (35%) than in the control group (18%). The CoULD group was less often employed and had lower scores on all measures of upper limb range of motion and hand grip. MSCs were associated with higher DASH scores and higher reported work demands. Disability was associated with female gender, more joints with limited range of motion, unemployment, and lower general and mental health. Factors associated with disability did not differ between groups.

Conclusions: MSCs are a frequent problem in young adults with major CoULD. To prevent or reduce MSC and disability, clinicians and researchers should be aware of the associated factors.

IMPLICATIONS FOR REHABILITATION

- The year prevalence of musculoskeletal complaints (MSCs) in those with major congenital upper limb differences (CoULD) was approximately double to that of the control group, implying a potential relationship between CoULD and MSCs.
- Rehabilitation professionals should develop personalized strategies to manage work demands in those with CoULD, considering the association between MSCs and higher reported work demands.
- Recognizing the impact of a negatively perceived body image on mental health, clinicians should integrate psychological counseling into rehabilitation treatments to support mental well-being and improve overall quality of life in those with CoULD.
- Rehabilitation professionals should educate individuals with CoULD about the potential associations between upper limb work demands, MSCs, and disability.

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Introduction

A missing, incomplete, or dysplastic hand or arm at birth is referred to as congenital upper limb anomaly, deficiency, or difference [1]. These congenital upper limb differences (CoULD) are rare, with a prevalence ranging between 10 and 30/10 000 [1–3]. Severity of CoULD can range from relatively minor differences such as clinodactyly to major transversal or longitudinal deficiencies, such as a cleft hand [4]. They can occur uni- or bilaterally, as part of a syndrome or together with other anomalies [3].

Since almost every daily activity requires a form of hand function, CoULD can impact functioning considerably. Therefore, children with CoULD are often followed-up by a pediatrician and/or rehabilitation physician during childhood, monitoring functioning, and managing limitations. As a result, most studies focusing on functioning with CoULD are limited to children. Interestingly, children and adolescents with CoULD appear to have almost normal participation in society, even though there can be considerable physical limitations or disability [5]. A Swedish survey showed that young adults with congenital limb differences (mostly unilateral transverse below elbow, but also longitudinal and lower extremities) function well [1]. They had normal employment and education compared to the general Swedish population and their general health was reported to be good to very good. However, a cross-sectional Norwegian study among adults with CoULD (transverse and longitudinal) reported a lower health-related quality of life and more bodily pain compared to able-bodied individuals [6]. In a Dutch survey, individuals with a transverse reduction deficiency experienced significantly more often

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musculoskeletal complaints (MSCs) compared to individuals without a transverse reduction deficiency (year prevalence of MSCs of 65% vs. 34%, respectively) [7]. Predictors for MSCs in individuals with transversal reduction deficiencies were: higher age, and lower mental and general health [7].

To our knowledge, no previous studies were conducted aimed at prevalence of MSCs and disability in adults with CoULD other than transverse type in the Netherlands. To provide better care for these individuals, more insight is needed on prevalence of MSCs and their effects. Generally, MSCs are related to a higher use of long-term healthcare services [8]. Prevention or early detection and treatment of MSCs may avoid disability, morbidity, and healthcare costs. Various biopsychosocial factors associated with MSCs have been identified in northwestern European working populations, such as increased psychosocial stress or lack of social support, awkward positions, and repetitive compensatory movements [9–13]. MSCs combined with the existing limitations can cause challenges in work participation and overall general and mental health [14]. Asymmetric postures, overuse and/or compensatory movements of the residual limb(s) of individuals with CoULD probably make them more prone to these psychological and biomechanical risk factors [15,16].

Most studies on congenital differences predominantly focus on transverse defects. In this way, specific information about other congenital limb anomalies is underreported. Our aim was to focus on "major" congenital limb differences besides transverse defects such as cleft hand or ulnar longitudinal deficiency.

The aims of this study were: (1) to determine the prevalence of MSCs in adults with major CoULD and in a group of individuals without CoULD (controls), and (2) to examine associations of MSCs and disability in the whole study with various biopsychosocial factors, such as joint mobility of the residual limb(s), reported limitations in functioning, work demands and general and mental health, in persons with CoULD and controls.

We hypothesize that individuals with major CoULD with a restricted function of one or both upper extremities are more likely to develop MSCs than able-bodied individuals.

Materials and methods

Design and setting

A questionnaire-based cross-sectional study comparing the study group with a control group was performed in 2018–2019. The 12 rehabilitation centers in the Netherlands that are treating individuals with CoULD were asked to provide a conclusive list of all individuals diagnosed with a major CoULD that had visited their Department of Rehabilitation Medicine. Three centers provided a list with possible participants: The Erasmus Medical Center in Rotterdam, De Hoogstraat Rehabilitation in Utrecht and the University Medical Center Groningen. These centers are the three main tertiary referral centers for patients with CoULD in the Netherlands. Ideally, all children with CoULD are referred to a rehabilitation physician by the pediatrician or general practitioner, and seen at least once in a rehabilitation center. However, it cannot be known for certain that all children are referred to a tertiary rehabilitation center.

The other nine centers could not provide a list with eligible participants, since they hardly treated individuals with CoULD or could not trace them in their registries.

Procedure

Between March and November 2018, eligible individuals were identified from the data registries of the mentioned centers. A control group matched on gender, age (±3 years), and education (lower = middle vocational education or lower; higher = vocational education or higher) was assembled between June 2019 and February 2020 among acquaintances of the researchers.

All eligible individuals were invited to fill out a questionnaire in November 2018. Fourteen days later, a reminder was sent to individuals who had not yet responded. A 10 euro gift voucher was sent to all participants who completed the questionnaire. In case of missing answers, the relevant questions were returned to the participant, with the request to answer these questions.

Inclusion criteria

We included individuals who had visited a rehabilitation center because of "major" CoULD (unilateral or bilateral), were 18 years or older on 1 November 2018 and had sufficient knowledge of the Dutch written language to fill in a questionnaire. As no definition of "minor" and "major" CoULD existed, we made a selection based on the Oberg, Manske, and Tonkin Classification of congenital anomalies of the hand and upper limb [15]. A diagnosis was qualified as "major" CoULD, when it was expected that it could limit the function of shoulder, elbow, wrist, and/or thumb, based on expert opinion of the last author, a rehabilitation physician, who has been working with individuals with CoULD for more than 20 years. The list of included diagnoses is shown in Appendix 1.

Exclusion criteria

We excluded individuals with solely minor congenital differences (Appendix 1): e.g., polydactyly, triphalangeal thumb, syndactyly, camptodactyly, thumb in palm deformity, clinodactyly, Kirner's deformity, synostose/symphalangism, congenital trigger digits, and reduction defects due to tumours, because of feasibility reasons and since we expected minimal to no disability in these individuals. We also excluded individuals with transversal reduction defects since these individuals had previously been investigated by our research group [7]. Although (congenital) tumours can cause major disability, we have excluded this group because (1) tumours with significant impact on upper extremity function mostly lead to amputation (e.g., transverse defect) [16]; (2) the malignancy itself and cancer treatment may cause additional health problems confounding the individuals' functioning.

Ethics

The local medical ethics committee decided that no formal ethical approval was necessary for this study (study number: 201500238).

Questionnaire

A modified version of a questionnaire previously developed for research in transverse upper limb reduction deficiency was used [7, 16]. It was assessed and pilot tested by field experts, fellow researchers, and a patient with a longitudinal upper limb reduction deficiency. The current questionnaire included the following elements.

CoULD-specific characteristics

Participants with CoULD were asked for the specific diagnosis/type of CoULD (if known), affected side, affected joints and/or bones and possible associated syndromes. Participants with CoULD were asked if they used a prosthesis.

The remainder of the questionnaire was identical for the CoULD and control group.

Participant characteristics

Date of birth, gender, civil status, education, hand dominancy, and possible previous surgery on the upper extremities were asked for. Also, employment status was asked for and, if employed, participants were asked to evaluate their work productivity.

Work productivity was assessed using the Quality-Quantity method (QQ-method), which was developed to measure the consequences of illness for work productivity. First participants are asked to indicate on a Visual Analogue Scale (VAS) scale from 1 to 10 how much work they actually performed during regular hours compared with the no-illness situation (quantity scale). Second, the quality of the work performed on a day is indicated on a VAS scale from 1 to 10 (quality scale). Multiplication of the quantity and quality components results in a QQ-score, which provides an indication of total performance, translating qualitative into quantitative losses (scoring range 1–100, a higher score indicating higher work quantity and quality) [17].

Physical work demands were assessed using the Revised Upper Extremity Work Demands (UEWD-R) Scale. This six-item questionnaire evaluates workload of the upper extremities such as: intensity, awkward body postures, and repetitive movements (scoring range 6–24, a higher score represents a higher workload of the upper extremities). The UEWD-R is valid and reliable; (intra-class correlation coefficient (ICC) = 0.79) [18]. Sick leave and sick leave hours were asked.

Musculoskeletal complaints

MSCs, complaints such as pain, stiffness, and tingling of the muscles, ligaments, bones, nerves, and/or joints, were asked for. The question was formulated in such a way that MSCs were made clearly distinguishable from complaints related to an accident, infection, sports injury, joint disease, stump pain, or phantom limb pain (see: Appendix 2). Point prevalence of MSCs was operationalized as the proportion of participants with MSCs in the past 4 weeks, and year prevalence of MSCs as the proportion of participants with MSCs in the past year (during a period of at least four consecutive weeks). If participants reported MSCs, they were asked to answer additional questions about the characteristics and treatment of their MSCs. For the location of MSCs, we used the image provided in the Dutch Musculoskeletal Questionnaire (DMQ) (see Appendix 3). On this image, participants can point out the exact location of their complaints. To reach a sufficient power for the statistical analyses, we combined the following regions: lower and upper back; shoulder and elbow; forearm and hand. DMQ is a questionnaire for the analysis of musculoskeletal workload and associated potential hazardous working conditions as well as musculoskeletal symptoms in worker populations [19].

Furthermore, intensity of bodily pain (not related to MSCs) during the last 4 weeks was assessed using a VAS (range 0–10), with a higher score indicating more pain.

Pain-related disability

All participants who experienced pain because of MSCs were asked to answer the Pain Disability Index (PDI). The PDI measures the degree daily life is disrupted by chronic pain [20]. Outcomes of the answers were summed up to a score ranging from 0 to 70. A higher score indicates more disability. The PDI is a valid and reliable tool (Cronbach's alpha = 0.65–0.89 and test–retest reliability (ICC) = 0.78) [21].

Disability

The Disability of Arm, Shoulder, and Hand Dutch Language Version (DASH-DLV) was included to assess disability related to hand function. The DASH is a 30-item, self-report questionnaire designed to measure physical function and symptoms in patients with musculoskeletal disorders of the upper limb. Each item can be scored on a five-point Likert scale (1: no difficulties, 5: unable). Outcomes of the answers were transformed to a score of 0–100. A higher score indicates more disability. The DASH-DLV is a reliable and valid instrument for assessing disability and symptoms (Cronbach's alpha = 0.95 and test–retest reliability, Pearson's r = 0.98) [22].

Health status

The participants' health status was measured using three subscales of the Dutch RAND 36-Item Health Survey: Bodily Pain, General Health, and Mental Health [23,24]. The RAND-36 is a set of self-reported quality-of-life measures, almost the same as the MOS SF-36 [23,24]. Item scores were transformed to a 0–100 scale, with a higher score indicating better perceived health or less pain. The RAND-36 for health status is a valid and sensitive measure (Cronbach's alpha = 0.81–0.88 and test–retest reliability (ICC) = 0.47–0.82) [25].

Comorbidity was categorized as: joint diseases (e.g., osteoarthritis, rheumatoid arthritis); circulatory diseases (e.g., heart-/vessel disease, high blood pressure); diabetes; pulmonary diseases; or other.

Appearance of the hand(s)

Participants were asked whether they found the appearance of their upper limb(s) disturbing, using a six-point Likert scale, where 1 reflected not disturbing at all and 6 reflected very disturbing [7].

Upper extremity function

Active joint movements of both limbs were assessed using self-developed rating scales. The movements were illustrated using pictures (see Appendix 3). Movements were abduction and external rotation of the shoulder, flexion and extension of the elbow, flexion and extension, ulnar and radial deviation of the wrist. The answers were dichotomized; with 0 representing: full range of motion possible; and 1: restricted range of motion. Absent joints were scored as 1. The total number of joints with restricted range of motion was summed up to determine the number of joints with limited range of motion (scoring range 0–8 per side).

Hand grip was assessed as follows: participants were asked if they could perform nine functional hand grips (opposition grip, lateral pinch, tip pinch, cylindrical grip, side-to-side grip, with and without use of the thumb, tripod pinch, hook grip, and spherical grip). These hand grips were illustrated using pictures (see: Appendix 3). For each handgrip, participants could answer on a four-point Likert-scale: 1: no difficulties in performing the movement, 2: can perform the movement, but with some difficulty, 3: cannot perform the entire movement, and 4: cannot perform the movement at all. A total grip score was calculated for left and right hand separately and converted to a percentage of the maximum possible score as follows: $100 - (\text{total score} - 9)/27) \times 100\%$. In addition, a total score for both limbs combined was calculated, ranging from 18 to 72, and converted to a percentage of the maximum possible score: $(\text{total score} - 18)/54) \times 100\%$. A higher score indicates a better hand grip. Validity of this method is not established yet.

Statistical analysis

Missing values for the DASH- and RAND-36-subscales were handled according to the manual of the respective questionnaires [26,27]. For the PDI and UEWD-R, a maximum of 1/7 and 1/6 missing items, respectively, were allowed and corrected for. Correction was done by imputing the mean of the answered items. In case of more missing items than allowed for, the score for that specific subscale was considered as a missing, and data-analysis was performed with listwise deletion. SPSS for Windows (version 23.0; SPSS Advanced Statistics, Chicago, IL) was used for data analyses. Categorical data were analysed with a Chi-square test and Cramer's *V* for determining the strength of association between variables and *r* as effect sizes were determined. For the year prevalence of MSCs, an odds ratio was determined. Continuous data were checked for normality by visual examination of QQ-plots and by interpretation of skewness and kurtosis statistics (both <1 was considered as a normal distribution). The normally distributed continuous variables were described with mean and standard deviation (SD) and analysed with an independent sample *t*-test. The test value (*t*), 95% confidence interval and effect size (by computing Cohen's *d*) were calculated. If data were not normally distributed, they were described with median and interquartile ranges (IQRs) and Mann–Whitney's *U*-test was used, and test statistic (*U*) and effect size were determined. Effect sizes (*r*) for Mann–Whitney's *U*-tests were calculated by dividing the *Z*-score by the square root of the study size. An effect size <0.3 was considered a small effect, 0.3–0.5 a medium effect, and >0.5 a large effect [28]. To measure the ordinal association between two measured quantities, Kendall's tau rank correlation coefficient (τ) was used. Variables associated with the presence of MSCs during the last year and with disability, represented by the DASH scores, were determined with multivariable logistic and linear regression analyses, respectively, using manual backwards selection method based on the largest *p* values. First, all independent variables from Tables 1–3 were univariately analyzed for their association with the outcome variables and those with a *p* value

Table 1. Characteristics of the studied population.

Characteristics	CoULD (n = 71)	Controls (n = 71)	Significance (p value)	Test statistics	Effect size	Magnitude of effect
Age	26.3 [21.5; 32.4]	27.2 [22.7; 32.2]	0.959	U = 2508.0	0.000	Nihil
Gender: male	29 (41%)	29 (41%)	1.000	χ^2 = 0.0	0.000	Nihil
Education						
Lower	6 (8%)	7 (10%)	0.957	χ^2 = 0.1	0.025	Small
Middle	43 (61%)	42 (60%)				
Higher	22 (31%)	22 (31%)				
Civil status			0.172	χ^2 = 1.8	0.115	Small
Single	46 (65%)	38 (53%)				
Living together	25 (35%)	33 (47%)				
Comorbidity	17 (24%)	10 (14%)	0.134	χ^2 = 2.2	0.126	Small
Employment						
Paid work	39 (55%)	49 (69%)	0.029	χ^2 = 4.7	0.183	Small
Working hours	32.0 [16.0; 38.0]	32.0 [21.0; 36.0]	0.860	U = 1907.5	−0.016	Small
Paid sick leave from job previous 4 weeks	5 (8%)	5 (7%)	0.834	χ^2 = 0.1	0.019	Small
Sick leave hours	24.0 [8.0; 24.0]	27.0 [16.0; 30.0]	0.690	U = 1000	−0.168	Small
No work because:						
Studying	23 (32%)	22 (31%)				
Hand/arm problems	1 (1%)	0 (0%)				
Disease (other than CoULD)	4 (6%)	0 (0%)				
Job searching	2 (3%)	0 (0%)				
Family care	2 (3%)	0 (0%)				
Upper limb characteristics						
Dominant hand						
Left	21 (29%)	4 (6%)				
Right	43 (61%)	65 (91%)				
Ambidexter	7 (10%)	2 (3%)				
Side of CoULD						
Left	23 (32%)					
Right	22 (31%)					
Bilateral	26 (37%)					
Surgery to hand or arm	46 (65%)	4 (6%)	<0.001	χ^2 = 54.4	0.619	Large
Hand grip score						
Left	88.9 [51.9; 100.0]	100.0 [100.0; 100.0]	<0.001	U = 934.5	−0.629	Large
Right	96.3 [51.9; 100.0]	100.0 [100.0; 100.0]	<0.001	U = 1161.0	−0.560	Large
Total	77.8 [57.4; 94.4]	100.0 [100.0; 100.0]	<0.001	U = 305.0	−0.816	Large
Limited active ROM						
Left shoulder	13 (18%)	2 (3%)	0.003	χ^2 = 9.0	0.252	Small
Right shoulder	13 (18%)	1 (1%)	0.001	χ^2 = 11.4	0.283	Small
Left elbow	16 (23%)	0 (0%)	<0.001	χ^2 = 18.0	0.356	Medium
Right elbow	13 (18%)	0 (0%)	<0.001	χ^2 = 14.3	0.317	Medium
Left wrist	24 (34%	0 (0%)	<0.001	χ^2 = 28.8	0.451	Medium
Right wrist	23 (32%)	0 (0%)	<0.001	χ^2 = 27.4	0.440	Medium
Number of limited joints	1.0 [0.0; 2.0]	0 [0.0; 0.0]	<0.001	U = 3849.0	0.569	Large

CoULD: congenital upper limb differences; ROM: range of motion; U: Mann–Whitney's U-test; χ^2: Chi-squared test.
The data are presented as: n (%) or median [interquartile range]. Hand grip score was assessed as follows: participants were asked if they could perform nine functional hand grips (opposition grip, lateral pinch, tip pinch, cylindrical grip, side-to-side grip (with and without use of the thumb), tripod pinch, hook grip, and spherical grip). These hand grips were illustrated using pictures. Each question could be answered on a four-point Likert scale: (1) no difficulties in performing the movement, (2) can perform the movement, but with some difficulty, (3) cannot perform the entire movement, and (4) cannot perform the movement at all. A total grip score was calculated for left and right hand separately and converted to a percentage of the maximum possible score as follows: 100 − (total score − 9)/27) × 100%. In addition, a total score for both limbs combined was calculated, ranging from 18 to 72, and converted to a percentage of the maximum possible score: (total score − 18)/54) × 100%. A higher score indicates a better hand grip. This method for scoring hand grip is not validated.

Table 2. Prevalence and characteristics of MSCs in CoULD and control group.

	CoULD (n = 71)	Controls (n = 71)	Significance (p value)	Test statistics	Effect size	Magnitude of effect
Point prevalence MSCs	24 (34%)	16 (23%)	0.136	χ^2 = 2.2	0.125	Small
Year prevalence MSCs	25 (35%)	13 (18%)	0.023	χ^2 = 5.2	0.191	Small
Location of complaints						
Neck	16 (23%)	10 (14%)	0.193	χ^2 = 1.7	0.109	Small
Upper and/or lower back	16 (23%)	10 (14%)	0.193	χ^2 = 1.7	0.109	Small
Shoulder and/or elbow left	12 (17%)	6 (9%)	0.130	χ^2 = 2.3	0.127	Small
Shoulder and/or elbow right	16 (23%)	10 (14%)	0.193	χ^2 = 1.7	0.109	Small
Arm and/or hand left	13 (18%)	5 (7%)	0.044	χ^2 = 4.1	0.169	Small
Lower arm and/or hand right	12 (17%)	5 (7%)	0.070	χ^2 = 3.3	0.152	Small
No. of regions with complaints	0.0 [0.0; 2.0]	0.0 [0.0; 1.0]	0.049	U = 2927.0	0.160	Small
Type of complaints						
Pain	26 (37%)	16 (23%)	0.811	χ^2 = 0.1	0.034	Small
Stiffness	14 (20%)	12 (17%)	0.260	χ^2 = 1.3	0.161	Small
Tingling	13 (18%)	2 (3%)	0.015	χ^2 = 5.9	0.347	Medium
Muscle weakness	9 (13%)	2 (3%)	0.111	χ^2 = 2.5	0.227	Small
Duration of complaints			0.541	χ^2 = 2.2	0.210	Small
<1 months	2 (3%)	3 (4%)				
1–3 months	4 (6%)	3 (4%)				
3–12 months	4 (6%)	4 (6%)				
≥12 months	20 (28%)	9 (13%)				
Healthcare use	18 (25%)	11 (15%)	0.884	χ^2 = 0.1	0.021	Small
Bodily pain score (VAS)	1.0 [0.0; 4.0]	0.0 [0.0; 1.0]	0.051	U = 2972.5	0.164	Small
PDI	10.0 [1.0; 29.0]	4.0 [1.0; 13.0]	0.383	U = 306.0	0.127	Small

MSCs: musculoskeletal complaints; CoULD: congenital upper limb differences; χ^2: Chi-squared test; U: Mann–Whitney's U-test; No.: number; PDI: Pain Disability Index. The data are presented as n (%) or median [interquartile range]. Data on PDI are based on: n = 47. Nine participants without MSCs accidentally filled out the PDI.

Table 3. Health, disability, and work outcomes in CoULD and control group during the last four weeks.

	CoULD (n = 71)	Controls (n = 71)	Significance (p value)	Test statistics	Effect size	Magnitude of effect
DASH	10.0 [3.3; 26.7]	1.7 [0.0; 3.3]	<0.001	U = 4120.0	0.553	Large
RAND-36 General health	75.0 [55.0; 95.0]	90.0 [70.0; 95.0]	0.007	U = 1865.5	−0.226	Small
RAND-36 Mental health	68.0 [60.0; 72.0]	72.0 [68.0; 76.0]	0.002	U = 1752.5	−0.265	Small
RAND-36 Bodily pain	20.4 [0.0; 42.9]	0.0 [0.0; 40.8]	0.027	U = 3038.5	0.186	Small
Disturbed by appearance arm/hand			<0.001	χ^2 = 44.7	0.561	Large
Never/rarely	41 (58%)	69 (97%)				
Sometimes	20 (28%)	2 (3%)				
Often/continuously	10 (14%)	0 (0%)				
Q-Q work productivity	100 [64.0; 100.0]	100.0 [100.0; 100.0]	0.003	U = 1560.0	−0.263	Small
UEWD-R	12.2 ± 4.1	12.3 ± 3.7	0.894	t = 0.1[a]	0.023	Small

CoULD: congenital upper limb differences; U: Mann–Whitney's U-test; χ^2: Chi-squared test; t = t value of Student's t-test; 95% CI: 95% confidence interval; DASH: Disability of Arm, Shoulder and Hand; RAND-36: RAND 36-Item Health Survey; Q-Q: quality quantity; UEWD-R: Revised Upper Extremity Work Demand Scale. The data are presented as: median [interquartile range] or n (%) or mean ± standard deviation. For variables where Chi-squared test is used, Cramer's V is given as effect size. For variables where Mann–Whitney's U-test is used, effect size is calculated by dividing the Z-score by the square root of the study size. For variables where Student's t-test is used, effect size is calculated by Cohen's d.
[a]95% CI = −1.47; 1.28.

<0.1 were selected for multivariable analyses. After main effects were determined in the multivariable analyses, interaction effects were explored and if $p \geq 0.1$ they were removed from the model. A p value ≤ 0.05 was considered statistically significant.

Results

In total, 176 surveys were sent to potential participants with CoULD. Seven surveys were returned to sender because of an incorrect address. Five participants denied participation and one survey was a duplicate, of which the first survey that was returned, was used. Seventy-one surveys (participation rate 41%) were eligible for analysis; the remaining individuals did not reply. Among the 105 individuals not included in the study, 52 were women and the median age of this group was 26.9 years [IQR: 22.8; 34.7]. In total, 73 surveys were sent to eligible participants of the matched control group. Seventy-one surveys were suitable for analysis (participation rate 97%). The characteristics of the study population (CoULD and controls) are presented in Table 1.

Of the participants with "major" CoULD, 32% was affected on the left side, 31% on the right side, and 37% was bilaterally affected. Based on the OMT-classification, they had the following upper limb differences: radial longitudinal deficiency (n = 13); symbrachydactyly (no forearm/arm involvement) (n = 12); ulnar longitudinal deficiency (n = 10); distal arthrogryposis (n = 9); symbrachydactyly (with forearm/arm involvement) (n = 8); cleft hand (n = 5); both ulnar and radial longitudinal deficiencies (n = 5); Poland syndrome (n = 3; all with longitudinal deficiencies of arm and/or hand); brachydactyly (n = 1); aberrant flexor/extensor/intrinsic muscle (n = 1); macrodactyly (n = 1), phocomelia (due to softenon; bilateral longitudinal deficiencies) (n = 1); not otherwise specified (n = 2).

The CoULD group was less often employed than the control group, had had surgery of the arm/hand more frequently and had lower scores on all measures of upper limb hand grip and active range of motion (Table 1). Three participants with CoULD use a (myoelectric) prosthesis.

Prevalence and characteristics of MSCs

Year prevalence of MSCs was significantly higher in the CoULD group (35%) than in the control group (18%); odds ratio 1.8 (Table 2). MSCs were most often located in the neck, upper, and/or lower back, and the right shoulder and/or elbow. Pain was the most frequent complaint in both groups. Tingling was more often reported by the CoULD group. In both groups, the duration of MSCs was often >1 year. Participants with CoULD did not differ significantly in MSC-related healthcare consumption compared to the control group. When comparing participants with CoULD with MSCs and controls with MSCs, no significant differences in PDI score were found.

Health, disability, and work outcomes

The CoULD group experienced more disability in upper limb function (higher DASH scores) and lower mental and general health (RAND-36 scores), compared to the control group (Table 3). They were also more often disturbed by the appearance of their hand and/or arm. Presence at work and work productivity (QQ-method) were significantly lower in the CoULD group. There were no significant differences between the CoULD group and controls regarding bodily pain or experienced upper extremity work demands (UEWD-R).

Factors associated with MSCs and disability

Multiple variables were associated with year prevalence of MSCs or disability (measured by the DASH score) in the total study population (Tables 4 and 5).

Multivariable logistic regression analyses in the total study population showed that the DASH score and UEWD-R score were associated with MSCs (Table 6), meaning that those with higher scores on the DASH and UEWD-R were more likely to experience MSCs.

Multivariable linear regression analyses showed that higher disability was related to female gender, being unemployed, low handgrip score, high number of joints with limited range of motion, and low general and mental health (Table 6). Furthermore, the combined effect of low mental and general health increased disability. Individuals with paid work experienced less disability due to limited range of motion of joints, compared to individuals without paid work.

Discussion

The year prevalence of MSCs in the CoULD group was about twice as high as in the control group. The hypothesis that these individuals are more likely to develop MSCs due to a restricted function of the upper extremities is not directly supported by this study since MSCs and restricted hand function were not associated in the logistic regression analysis. However, we found an indirect association. A more severely restricted hand function reflected by a higher number of joints with limited range of motion (large effect size) and a lower hand grip strength (large effect size), were associated with disability (DASH scores, which were much higher in the CoULD group), while disability was found to be associated with presence of MSCs. MSCs were also associated with higher reported work demands of the upper extremity. Disability was

Table 4. Factors associated with year prevalence of MSCs in the studied population.

	Individuals with MSCs (n = 38)	Individuals without MSCs (n = 104)	Significance (p value)	Test statistics	Effect size	Magnitude of effect
Presence of CoULD	25 (66%)	46 (44%)	0.023	χ^2 = 5.2	0.191	Small
Age	29.2 [24.9; 33.3]	25.3 [21.5; 31.8]	0.046	U = 2409.0	0.100	Small
Male gender	15 (40%)	43 (41%)	0.841	χ^2 = 0.1	0.170	Small
Civil status			0.339	χ^2 = 0.9	0.080	Small
Single	20 (53%)	64 (61%)				
Living together	18 (47%)	40 (39%)				
Education			0.603	χ^2 = 1.0	0.117	Small
Lower	5 (13%)	8 (7%)				
Middle	22 (58%)	63 (61%)				
Higher	11 (29%)	33 (32%)				
Surgery of arm and/or hand	12 (32%)	38 (36%)	0.584	χ^2 = 0.3	0.046	Small
Comorbidity	10 (26%)	17 (16%)	0.180	χ^2 = 1.8	0.112	Small
Hand grip score	82.4 [50.0; 100.0]	100.0 [88.9; 100.0]	<0.001	U = 1168.0	−0.336	Medium
Limited active ROM						
Left shoulder	8 (21%)	7 (7%)	0.014	χ^2 = 6.0	0.206	Small
Right shoulder	8 (21%)	6 (6%)	0.007	χ^2 = 7.3	0.227	Small
Left elbow	8 (21%)	8 (8%)	0.026	χ^2 = 4.9	0.187	Small
Right elbow	8 (21%)	5 (5%)	0.003	χ^2 = 8.8	0.249	Small
Left wrist	11 (29%)	13 (13%)	0.021	χ^2 = 5.4	0.194	Small
Right wrist	11 (29%)	12 (12%)	0.013	χ^2 = 6.2	0.209	Small
Number of joints with limited ROM	0.5 [0.0; 2.0]	0.0 [0.0; 0.0]	0.001	U = 2551.0	0.278	Small
DASH	16.7 [2.5; 33.3]	2.5 [0.0; 7.5]	<0.001	U = 2940.5	0.376	Medium
RAND-36 General health	68.0 [50.0; 95.0]	85.0 [70.0; 95.0]	0.006	U = 1379.0	−0.232	Small
RAND-36 Mental health	64.0 [52.0; 72.0]	72.0 [68.0; 76.0]	0.002	U = 1312.0	−3.088	Large
Paid work	29 (76%)	69 (66%)	0.255	χ^2 = 1.3	0.095	Small
Work hours	30.0 [24.0; 36.5]	32.0 [19.0; 36.5]	0.282	U = 1568.0	0.025	Small
Sick leave	3 (9%)	7 (7%)	0.713	χ^2 = 0.1	0.031	Small
Sick leave hours	24.0 [8.0; 30.0]	24.0 [8.0; 31.0]	0.817	U = 9500.0	0.258	Small
Q-Q evaluation	95.0 [49.0; 100.0]	100.0 [90.0; 100.0]	0.007	U = 1196.0	−0.237	Small
UEWD-R	13.5 ± 4.9	11.8 ± 3.4	0.080	t = 1.8	0.403	Medium

MSCs: musculoskeletal complaints; CoULD: congenital upper limb differences; CI: confidence interval; U: Mann–Whitney's U-test; χ^2: Chi-squared test; t: t value of Student's t-test; 95% CI: 95% confidence interval; M.D.: mean difference; ROM: range of motion; No.: number; DASH: Disability of Arm, Shoulder and Hand; RAND-36: RAND 36-Item Health Survey; Q-Q: quality quantity; UEWD-R: Revised Upper Extremity Work Demands Scale.
The data are presented as: n (%) or median [interquartile range] or mean ± standard deviation. For variables where Chi-squared test is used, Cramer's V is given as effect size. For variables where Mann–Whitney's U-test is used, effect size is calculated by dividing the Z-score by the square root of the study size.

Table 5. Univariate analyses of factors associated with disability measured by the DASH score within the total study population.

	Significance (*p* value)	Test statistics	Effect size	Magnitude of effect
CoULD	<0.001	U = 2940.5	0.376	Medium
Age	0.871	τ = 0.010		
Gender	0.004	U = 3129.5	0.243	Small
Civil status	0.121	U = 2065.5	−0.130	Small
Education	0.412	τ = −0.057		
Surgery	<0.001	U = 3146.5	0.306	Medium
Comorbidity	0.001	U = 2181.5	0.277	Small
Number of joints with limited ROM	<0.001	τ = 0.445		
Hand grip score	<0.001	τ = −0.543		
RAND-36 Mental health	<0.001	τ = −0.295		
RAND-36 General health	<0.001	τ = −0.333		
Paid work	0.072	U = 1751.5	−0.151	Small
Working hours/ week	0.082	τ = −0.113		
Sick leave last 4 weeks	0.311	U = 691.0	0.090	Small
Hours of sick leave	0.583	τ = −0.141		

CoULD: congenital upper limb differences; τ: Kendall's Tau; *U*: Mann–Whitney's *U*-test; No.: number; DASH: Disability of Arm, Shoulder and Hand; RAND-36: RAND 36-Item Health Survey.
For variables where Chi-squared test is used, Cramer's *V* is given as effect size. For variables where Mann–Whitney's *U*-test is used, effect size is calculated by dividing the *Z*-score by the square root of the study size.

also associated with female gender, employment, hand function, and lower general and mental health.

Besides differences, there were also similarities between the groups. For instance, there were no significant differences between the CoULD group and controls on: working hours, sick leave, and experienced work demands.

MSCs and disability

MSCs and age

The significantly higher year prevalence of MSCs in adults with major CoULD compared to a group of able-bodied people is in line with earlier findings in individuals with congenital transverse differences [7]. Interestingly, the year prevalence of MSCs in the CoULD group (35%) was similar to the year prevalence of MSCs in the general Dutch population found previously (37%) [29], while the prevalence in current control group was much lower. This difference is probably related to the young age of our study population. In a study among individuals with an upper limb transverse reduction deficiency or amputation, higher age was found to be associated with higher risk on MSCs [7]. In the current study, the group of individuals who experienced MSCs were significantly older than the group who did not experience MSCs (difference in median age: 4 years).

Characteristics of MSCs, and its relation with health and work

Pain was the most reported type of MSC and MSCs were most frequently located at the neck, back and right shoulder and/or elbow. Individuals with CoULD were more often ambidextrous or left-handed, compared to the control group. This finding suggests that, in order to meet functional demands and compensate for disability of the affected limb, presence of CoULD influences hand dominance?

Table 6. Results of logistic regression analyses to predict year prevalence of MSCs and linear regression analyses to predict disability measured with the DASH score.

	B	S.E.	Significance (*p* value)	Odds ratio	95% CI
MSCs[a]					
DASH	0.082	0.021	<0.001	1.085	1.043; 1.130
UEWD-R	0.109	0.058	0.060	1.115	0.995; 1.250
Constant[b]	−3.232	0.829	<0.001	0.039	
DASH[c]					
Gender	4.541	1.572	0.005		1.433; 7.650
Paid work	−5.152	1.686	0.003		−8.486; −1.817
Hand grip score	−0.299	0.049	<0.001		−0.397; −0.202
RAND-36 Mental health	−0.173	0.077	0.026		−0.325; −0.020
RAND-36 General health	−0.164	0.042	<0.001		−0.247; −0.081
Number of joints with limited ROM	2.737	0.688	<0.001		1.377; 4.098
RAND-36 Mental health × RAND-36 General health	0.009	0.003	0.007		0.002; 0.015
Paid work × number of joints with limited ROM	−4.627	0.975	<0.001		−6.555; −2.698
Constant[d]	43.905	5.610	<0.001		32.809; 55.001

B: regression coefficient; SE: standard error; CI: confidence interval; MSCs: musculoskeletal complaints; DASH: Disability of Arm; Shoulder and Hand; UEWD-R: Revised Upper Extremity Work Demands Scale; RAND-36: RAND 36-Item Health Survey; ROM: range of motion.
The results of the linear regression analysis indicated that women had a higher score (4.5) than men and that per joint with a restricted mobility the DASH score was 2.7 points higher. Having paid work was associated with lower DASH scores (5.1) and per point grip score the DASH score was 0.3 points lower. Additionally, higher mental health scores and general health scores were associated with lower DASH scores (0.2 per point on the RAND-36). The interaction term indicates that the combined effect of mental health scores and general health scores was somewhat higher: 0.009 per point of the product of mental and general health. Additionally, the interaction term between paid work and number of joints with a limited range of motion indicates that if you have paid work, the DASH score is 4.6 point lower per affected joint. For the logistic regression analysis, the odds ratio is the effect size, in the linear regression analysis, the regression coefficients (*B*) is the effect sizes.
[a]Eighty-one percent was correctly predicted with the results of the logistic regression analysis.
[b]The constant reflects the odds ratio of having MSCs for an individual with the following features: DASH-score is 0 (indicating no disability) and UEWD-R is 6 (indicating low physical workload for the upper extremities).
[c]R^2 of the total model was 0.737; residuals were normally distributed.
[d]The constant is the DASH score for an individual with the following features: male gender, no paid work, hand grip score is 0, no limited range of motion of joints and RAND-36 mental and general health score are both 0.

Despite their young age, the majority of the participants with MSCs reported chronic complaints, lasting more than one year. Combined with the relatively high prevalence, this chronicity also exposes the potential impact of MSCs in these individuals and on society. Most participants with MSCs, both those with CoULD as well as the controls, sought care through some type of healthcare professional, which reflects a potential high burden for the healthcare system. Furthermore, work productivity was negatively affected by the presence of MSCs.

Participants with CoULD mentioned more bodily locations affected by MSCs, compared to the control group. Nevertheless, disability measured with the PDI did not differ between both groups.

Disability

Regression analyses showed no main effect of presence of CoULD on disability, indicating that this relatively young population seems to function similar as able-bodied individuals, which conforms earlier findings in Norway (median age in this study: 39 years) [30]. However, it is important to note that the Norwegian study included more transverse than longitudinal defects (101 vs. 16). Whether these findings also hold for older individuals with CoULD could not be confirmed, since we did not include many elderly. Disability related to upper limb function, measured with the DASH, was significantly higher in the CoULD population than in the controls. The association between CoULD and physical capabilities was investigated by examining hand grip function and joint mobility. Individuals with CoULD had lower grip scores than controls and had a higher number of joints with limited range of motion; note that only three controls (without CoULD) had a limited range of motion. These limitations in physical capabilities may explain why individuals with CoULD experience higher disability in upper limb functioning. This finding is in line with finding among subjects with arthrogryposis, in which disability and pain is frequently present [31,32].

MSCs were not only associated with higher upper limb related disability, but also with higher upper limb related work demands. Those with CoULD had less often paid work and lower work productivity (as measured with the QQ-method). In the general Dutch population, work disability among men and women is similar, although younger women in the Netherlands seem to be more prone to work disability due to mental problems [30, 33]. Given the young mean age of our population, this could be an explanation for our finding that only female gender was associated with disability.

Limited physical capabilities of the upper limbs may lead to compensatory movements and awkward postures, which may cause overuse and MSCs. However, the opposite may also be applicable: presence of MSCs may lead to compensatory movements and awkward postures in order to relieve the painful body structures. High upper extremity work demands and disability may inflict MSCs in the same manner, but MSCs may also lead to an increase in the experienced level of work demands and disability. Due to the cross-sectional design of our study, it is not possible to unravel any causal relationships regarding work demands, disability and MSCs. However, clinicians should be aware of the fact that upper limb work demands are associated with MSCs and disability and should therefore advise their patients accordingly, preferably in an early phase of their life.

Participants with CoULD had more bodily pain and lower general and mental health than the control group. With higher perceived disability, general and mental health were perceived to be lower, which is in line with previous research among individuals with transverse reduction differences and acquired amputation of the upper extremity in the Netherlands and CoULD in Norway [8, 19, 34].

Additionally, participants with CoULD were more often disturbed by the appearance of their hand and/or arm. A negatively perceived body image due to congenital differences was associated with lower mental health and quality of life in adolescents. Many of those adolescents develop adequate coping strategies but it is plausible that some adults still suffer from a negatively perceived body image [35,36]. The attention of clinicians and future research should be focused on mental and general health; since these factors may potentially be influenced by an intervention. Psychological counseling focusing at mental well-being and perceived body image should therefore be part of the rehabilitation treatment in individuals with CoULD.

Strengths and limitations of the study

This is one of few studies on functioning of adults with CoULD [5–7, 14, 30, 34, 37,38]. Given the very low incidence of CoULD, the sample size of the study group is respectable, but the CoULD diagnoses of this group were heterogeneous, making it more difficult to generalize results. The participation rate was sufficient and similar to studies using postal questionnaires [39]. Matched controls were used to eliminate confounding by gender, age, and education. The matching was successfully established, as no differences for these confounders were found when comparing participants with CoULD and controls. The available registry of people with CoULD at the UMCG went back until the 1970s, providing a large age range among possible participants. In contrast, the registry of the other participating centers went back until the 2000s, which led to relatively young potential participants.

It appeared to be hard to identify individuals with CoULD of higher age, due to limitations in the registries of the participating rehabilitation centers. The reported prevalence is probably an underestimation of the true prevalence in all adults with CoULD, due to our young study population (75% of participants had an age <33 years). MSCs are known to be age-related with an increase in prevalence around the fourth decade [7, 29, 40]. Preferably, our study should be repeated in a CoULD population of higher age, since that would provide more insight into the consequences of MSCs in this population. Development of MSCs over time requires a longitudinal study.

Our inclusion criteria might have led to a selection bias. The diagnoses that were selected as "major" CoULD were based on expert opinion. There is no internationally accepted classification regarding the severity of CoULD. In hindsight, two diagnoses should not have been qualified as "major" CoULD: "aberrant flexor/extensor/intrinsic muscle" and "brachydactyly". We only included two patients with these respective conditions. When repeating the analyses without these two cases, similar outcomes were found (outcomes are available on request to the corresponding author). All rehabilitation centers in the Netherlands were contacted to recruit people with these rare conditions. However, if potential participants never visited a rehabilitation center they could not be contacted for this research. This phenomenon may have led to a selection bias but we expect that those who have never visited a rehabilitation center are mostly the individuals with minor CoULD. Furthermore, bias could have occurred by using acquaintances, instead of random samples of individuals, as matched control subjects. Some types of CoULD can cause other major disabilities, for instance to the lower extremities; these could possibly have influenced functioning and workload of the upper extremities [1]. The use of self-developed rating scales to measure the active range of motion of the joints and hand grips may have compromised validity and reliability. Physical examination instead of asking after active range of joint motion and hand grips would have been preferable, but was not feasible within this study due to travel distances and a lack of resources.

Including all studied individuals in the multivariable regression analysis allows to analyse whether MSCs and disability are related to CoULD itself, or to its consequences, such as limited joint mobility. Furthermore, it allows to control for confounding variables, for example, mental health status, and the occurrence of mediation. The included univariate analyses explain the associations further. Last, including both groups in the analysis increases the statistical power.

For example, in the regression analysis to statistically predict disability (depicted by DASH scores), we first entered CoULD (yes/no) and a significant association was found. When the variable "number of joints with restricted mobility" is entered, the association disappears. This phenomenon is called mediation. It means that the association between CoULD and higher DAHS scores is completely explained by the number of restricted joints and that the association is similar for CoULD as well as controls.

Conclusions

The year prevalence of MSCs in adults with major CoULD was twice as high as the year prevalence of MSCs in a matched group of able-bodied people. MSCs were associated with higher upper limb related disability and higher reported work demands of the upper extremity. Disability was associated with female gender, more joints with limited range of motion, worse hand grip, no employment, and lower general and mental health.

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References

[1] Wilcox WR, Coulter CP, Schmitz ML. Congenital limb deficiency disorders. Clin Perinatol. 2015;42(2):281–300, viii. doi: 10.1016/j.clp.2015.02.004.
[2] Vasluian E, van der Sluis CK, van Essen AJ, et al. Birth prevalence for congenital limb defects in the Northern Netherlands: a 30-year population-based study. BMC Musculoskelet Disord. 2013;14:323.
[3] Goldfarb CA, Shaw N, Steffen JA, et al. The prevalence of congenital hand and upper extremity anomalies based upon the New York Congenital Malformations Registry. J Pediatr Orthop. 2017;37(2):144–148. doi: 10.1097/BPO.0000000000000748.
[4] Goldfarb CA, Ezaki M, Wall LB, et al. The Oberg–Manske–Tonkin (OMT) classification of congenital upper extremities: update for 2020. J Hand Surg. 2020;45(6):542–547.
[5] Holtslag I, Van Wijk I, Hartog H, et al. Long-term functional outcome of patients with longitudinal radial deficiency: cross-sectional evaluation of function, activity and participation. Disabil Rehabil. 2013;35(16):1401–1407. doi: 10.3109/09638288.2012.737086.
[6] Johansen H, Østlie K, Andersen LØ, et al. Health-related quality of life in adults with congenital unilateral upper limb deficiency in Norway. A cross-sectional study. Disabil Rehabil. 2016;38(23):2305–2314. doi: 10.3109/09638288.2015.1129450.
[7] Postema SG, Bongers RM, Brouwers MA, et al. Musculoskeletal complaints in transverse upper limb reduction deficiency and amputation in The Netherlands: prevalence, predictors, and effect on health. Arch Phys Med Rehabil. 2016;97(7):1137–1145. doi: 10.1016/j.apmr.2016.01.031.
[8] Hartvigsen J, Hestbaek L, Davidsen M, et al. Self-reported musculoskeletal pain predicts long-term increase in general health care use: a population-based cohort study with 20-year follow-up. Scand J Public Health. 2014;42(7):698–704. doi: 10.1177/1403494814542263.
[9] IJzelenberg W, Molenaar D, Burdorf A. Different risk factors for musculoskeletal complaints and musculoskeletal sickness absence. Scand J Work Environ Health. 2004;30(1):56–63. doi: 10.5271/sjweh.765.
[10] van Rijn RM, Huisstede BM, Koes BW, et al. Associations between work-related factors and specific disorders of the shoulder – a systematic review of the literature. Scand J Work Environ Health. 2010;36(3):189–201. doi: 10.5271/sjweh.2895.
[11] Feveile H, Jensen C, Burr H. Risk factors for neck-shoulder and wrist-hand symptoms in a 5-year follow-up study of 3,990 employees in Denmark. Int Arch Occup Environ Health. 2002;75(4):243–251. doi: 10.1007/s00420-001-0298-9.
[12] Yassi A. Repetitive strain injuries. Lancet. 1997;349(9056):943–947. doi: 10.1016/S0140-6736(96)07221-2.
[13] Reneman MF, Soer R, Gerrits EHJ. Basis for an FCE methodology for patients with work-related upper limb disorders. J Occup Rehabil. 2005;15(3):353–363. doi: 10.1007/s10926-005-5942-x.
[14] Johansen H, Bathen T, Andersen LØ, et al. Education and work participation among adults with congenital unilateral upper limb deficiency in Norway: a cross-sectional study. PLOS One. 2018;13(12):e0207846. doi: 10.1371/journal.pone.0207846.
[15] Tonkin MA. Classification of congenital anomalies of the hand and upper limb. J Hand Surg Eur. 2017;42(5):448–456.
[16] Duran-Moreno J, Kontogeorgakos V, Koumarianou A. Soft tissue sarcomas of the upper extremities: maximizing treatment opportunities and outcomes. Oncol Lett. 2019;18(3):2179–2191. doi: 10.3892/ol.2019.10575.
[17] Brouwer WBF, Koopmanschap MA, Rutten FFH. Productivity losses without absence: measurement validation and empirical evidence. Health Policy. 1999;48(1):13–27. doi: 10.1016/s0168-8510(99)00028-7.
[18] Cavalini MA, Berduszek RJ, van der Sluis CK. Construct validity and test–retest reliability of the Revised Upper Extremity Work Demands (UEWD-R) Scale. Occup Environ Med. 2017;74(10):763–768. doi: 10.1136/oemed-2017-104370.
[19] Hildebrandt VH, Bongers PM, Dijk FJH, et al. Dutch Musculoskeletal Questionnaire: description and basic. Methods. 2001;44(12):1038–1055. doi: 10.1080/00140130110087437.
[20] Tait RC, Chibnall JT, Krause S. The Pain Disability Index: psychometric properties. Pain. 1990;40(2):171–182. doi: 10.1016/0304-3959(90)90068-O.
[21] Soer R, Köke AJA, Vroomen PCAJ, et al. Extensive validation of the Pain Disability Index in 3 groups of patients with musculoskeletal pain. Spine (Phila Pa 1976). 2013;38(9):E562–E568. doi: 10.1097/BRS.0b013e31828af21f.
[22] Veehof MM, Sleegers EJA, van Veldhoven NHMJ, et al. Psychometric qualities of the Dutch language version of the Disabilities of the Arm, Shoulder, and Hand Questionnaire (DASH-DLV). J Hand Ther. 2002;15(4):347–354. doi: 10.1016/s0894-1130(02)80006-0.
[23] Ware JE, Sherbourne CD. The MOS 36-item Short-Form Health Survey (SF-36). Med Care. 1992;30(6):473–483. doi: 10.1097/00005650-199206000-00002.
[24] Aaronson NK, Muller M, Cohen PDA, et al. Translation, validation, and norming of the Dutch language version of the SF-36 Health Survey in community and chronic disease populations. J Clin Epidemiol. 1998;51(11):1055–1068. doi: 10.1016/s0895-4356(98)00097-3.
[25] van der Zee K, Sanderman R. Het meten van de algemene gezondheidstoestand met de RAND-36. Noordelijk Centrum voor Gezondheidsvraagstukken, reeks meetinstrumenten. 1993;3:1–28.

[26] Kennedy CA, Beaton DE, Solway S, et al. Disabilities of the Arm, Shoulder and Hand (DASH). The DASH and QuickDASH outcome measure user's manual. 3rd edition, Toronto, Canada; 2011.
[27] Van der Zee K, Sanderman R. Rand-36. Vol. 28. Groningen, The Netherlands: Northern Centre for Health Care Research, University of Groningen; 1993.
[28] Field A. Discovering statistics using IBM SPSS statistics. 4th ed.; 2013.
[29] Picavet HSJ, Schouten JSAG. Musculoskeletal pain in The Netherlands: prevalences, consequences and risk groups, the DMC(3)-study. Pain. 2003;102(1–2):167–178. doi: 10.1016/s0304-3959(02)00372-x.
[30] Johansen H, Østlie K, Andersen LØ, et al. Adults with congenital limb deficiency in Norway: demographic and clinical features, pain and the use of health care and welfare services. A cross-sectional study. Disabil Rehabil. 2015;37(22):2076–2082. doi: 10.3109/09638288.2014.998781.
[31] Dai S, Dieterich K, Jaeger M, et al. Disability in adults with arthrogryposis is severe, partly invisible, and varies by genotype. Neurology. 2018;90(18):e1596–e1604. doi: 10.1212/WNL.0000000000005418.
[32] Cirillo A, Collins J, Sawatzky B, et al. Pain among children and adults living with arthrogryposis multiplex congenita: a scoping review. Am J Med Genet C Semin Med Genet. 2019;181(3):436–453. doi: 10.1002/ajmg.c.31725.
[33] Louwerse I, Huysmans MA, Van Rijssen HJ, et al. Characteristics of individuals receiving disability benefits in The Netherlands and predictors of leaving the disability benefit scheme: a retrospective cohort study with five-year follow-up. BMC Public Health. 2018;18(1):157. doi: 10.1186/s12889-018-5068-7.
[34] Datta D, Selvarajah K, Davey N. Functional outcome of patients with proximal upper limb deficiency – acquired and congenital. Clin Rehabil. 2004; 18(2):172–177. doi: 10.1191/0269215504cr716oa.
[35] Miller R, Samarendra H, Hotton M. A systematic review of the use of psychological assessment tools in congenital upper limb anomaly management. J Hand Ther. 2020;33(1):2–12.e1. doi: 10.1016/j.jht.2018.11.001.
[36] Franzblau LE, Chung KC, Carlozzi N, et al. Coping with congenital hand differences. Plast Reconstr Surg. 2015;135(4):1067–1075. doi: 10.1097/PRS.0000000000001047.
[37] Bae DS, Canizares MF, Miller PE, et al. Functional impact of congenital hand differences: early results from the Congenital Upper Limb Differences (CoULD) registry. J Hand Surg Am. 2018;43(4):321–330. doi: 10.1016/j.jhsa.2017.10.006.
[38] Sjöberg L, Nilsagård Y, Fredriksson C. Life situation of adults with congenital limb reduction deficiency in Sweden. Disabil Rehabil. 2014;36(18):1562–1571. doi: 10.3109/09638288.2013.854839.
[39] Cook JV, Dickinson HO, Eccles MP. Response rates in postal surveys of healthcare professionals between 1996 and 2005: an observational study. BMC Health Serv Res. 2009;9(1):160. doi: 10.1186/1472-6963-9-160.
[40] Bouma SE, Postema SG, Bongers RM, et al. Musculoskeletal complaints in individuals with finger or partial hand amputations in The Netherlands: a cross-sectional study. Disabil Rehabil. 2018;40(10):1146–1153. doi: 10.1080/09638288.2017.1289418.