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Abstract—**Facioscapulohumeral Dystrophy (FSHD) is a progressive muscle-wasting disease which leads to a decline in upper extremity functionality. Although the scapulohumeral joint's stability and functionality are affected, evidence on the synergetic control of the shoulder muscles in FSHD individuals is still lacking. The aim of this study is to understand the neuromuscular changes in shoulder muscle control in people with FSHD. Upper arm kinematics and electromyograms (EMG) of eight upper extremity muscles were recorded during shoulder abduction-adduction and flexion-extension tasks in eleven participants with FSHD and eleven healthy participants. Normalized muscle activities were extracted from EMG signals. Non-negative matrix factorization was used to compute muscle synergies. Maximum muscle activities were compared using non-parametric analysis of variance . Similarities between synergies were also calculated using correlation. The Biceps Brachii was significantly more active in the FSHD group (25±2%) while Trapezius Ascendens and Serratus Anterior were less active (32±7% and 39±4% respectively). Muscle synergy weights were altered in FSHD individuals and showed greater diversity while controls mostly used one synergy for both tasks. The decreased activity by selected scapula rotator muscles and muscle synergy weight alterations show that neuromuscular control of the scapulohumeral joint is less consistent in people with FSHD compared to healthy participants. Assessments of muscle coordination strategies can be used to evaluate motor output variability and assist in management of the disease.**

Index Terms—: **FSHD, humeral elevation, motor control, muscular dystrophy, muscle synergies, scapula rotation.**

I. INTRODUCTION

FACIOSCAPULOHUMERAL Dystrophy (FSHD) is characterized by progressive muscle wasting which primarily affects the face and shoulder area [1], [2]. Muscle quality decreases due to fat infiltration, but is weakly correlated with age where age onset varies greatly [2], [3]. Commonly occurring body impairments and functional limitations include scapular winging, joint instability, and a decline in upper extremity functionality [4], [5], [6], [7], [8]. In a questionnaire-based survey, reaching and lifting objects above shoulder level were reported as "most limited" activities by 45% of FSHD participants [6]. Relative surface area, as a measure of the reachable workspace, decreases by 23 to 87% depending on the level of strength loss, in people with FSHD [9], [10]. Muscles attaching to the scapula are the most affected, with the Trapezius and Serratus Anterior muscles becoming atrophied and showing fat infiltration in more than 85% of individuals with FSHD [11]. These losses in tissue quantity and quality become evident at the earliest stages of the disease [11], [12] and translate into a diminished strength of the scapular rotator muscles. In turn, this limited muscle function could result in incomplete rotation and stabilization of the scapula.

Electromyographic assessments of muscle function can provide insight in the muscle activation strategies used for scapular stabilization and mobilization in people with FSHD. Previous research has shown an approximately twice as high muscle activity in FSHD participants compared to healthy individuals for the Deltoid, Trapezius Descendens, and synergist Biceps muscles during reaching tasks [13]. The increased activity of selected shoulder muscles can be postulated to compensate for the loss of strength, with scapular mobilization possibly affected as a result. In healthy individuals, scapular mobilization and stability are necessary during humeral elevation, particularly above shoulder level [14], [15], [16]. At present however, the way in which scapular rotator and humeral elevator muscles are coordinated by FSHD individuals during daily tasks is still unclear. The extent of these alterations that are known to occur in other diseases affecting the shoulder, including stroke, multiple sclerosis, and shoulder impingement [17], [18], [19], [20], [21], [22], indicate that the neuromuscular output can be affected by the disease.

Muscle synergy analysis can be used to reveal alterations in the coordination of groups of muscles. In healthy individuals the central nervous system activates muscles in groups, as a neural strategy to simplify the control of multiple degrees of freedom [23]. These group activations, commonly called muscle synergies, can be described by the relative contribution of each muscle (weights) during a common time-dependent

activation command (coefficients) [24]. Muscle synergy analysis of the upper extremity in people post-stroke has revealed alterations in the shoulder muscle synergies during isometric force generation [25] and dynamic tasks [26]. A high similarity between affected and unaffected arm muscle synergies was shown in a variety of daily activities, together with the presence of compensatory strategies by Trapezius and Pectoralis muscles during reaching tasks [25], [27], [28]. In people with FSHD, however, it is unknown how muscle synergies change during the execution of upper extremity daily tasks. Understanding the neuromuscular output can help reveal how the disease-resulting changes in kinematics are underlined by muscular changes, with implications for the long-term management of the condition.

This study concentrates on planar humeral elevation tasks to understand the neuromuscular changes affecting the shoulder muscles, including muscles responsible for scapula rotation and stabilization, in people with FSHD compared to healthy individuals. We hypothesized that in people with FSHD the maximum activity of prime movers of humerus and scapula and of synergist muscles would be higher compared to healthy individuals. Secondly, we also hypothesized that muscle synergies would show alterations in people with FSHD, reflecting the increase in maximum activity, mainly in synergy weights. The second hypothesis was tested to investigate whether the known shoulder mobility limitations in people with FSHD would affect the muscle synergies.

II. METHODS

A. Participants

Eleven healthy control participants (5M/6F, 55±14ys, 175±7cm, 69±8kg, 11Right-Dominant (RD)) and eleven participants with FSHD (6M/5F, 54±15ys, 177±11cm, 78±21kg, 2LD/9RD) were included in this study. Healthy participants were informed by advertisement flyers located at University Medical Center Groningen. People with FSHD were informed about the study through the Dutch Association for Neuromuscular Diseases (Spierziekten Nederland, Baarn, NL). Healthy and participants with FSHD were included in this study
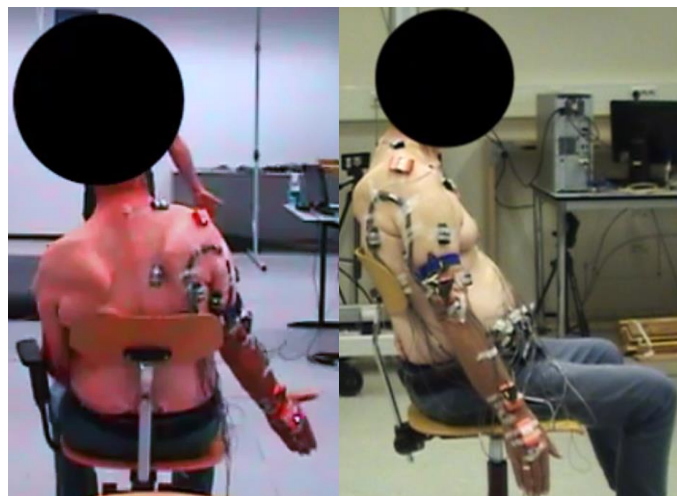
if they were aged between 18-75 years, able to read and understand Dutch, and able to give written informed consent. Additional criteria for people with FSHD were the ability to transfer from wheelchair to chair with side- and lower back-rest, and a Brooke scale score of 3 or 4. Healthy participants were excluded if they were diagnosed with pathologies that could interfere with the measurement results, had a presence of pain in the shoulder, a history of severe trauma of the shoulder within the previous two years (e.g. fracture, luxation). Participants with FSHD were excluded if they had comorbidities that could interfere with the measurement results, previous surgery on the right shoulder, extrinsic causes of shoulder pain, a history of severe trauma, or were unable to elevate the right arm above 30°. Age, gender, hand-dominance, body height, and body mass were also recorded. The central Medical Ethical Committee of University Medical Center Groningen approved the study (NL55711.042.15), which was carried out in accordance with the guidelines of the Helsinki protocol. Participants were informed about the procedure beforehand and provided written informed consent.

B. Movement tasks

The participants were positioned in a chair with a left side-rest and lower back-rest and with the seat height adjusted to achieve a knee flexion angle of 90°. Participants received detailed instructions prior to the execution of each task regarding the movement. For the shoulder abduction-adduction task (SAA), the right arm was first positioned downward with the elbow straight and the hand palm facing forward (Fig. 1). The movement consisted of lifting the arm as far as possible in the coronal plane and bringing it back to the start position while keeping the trunk and elbow straight, with the hand palm facing forward. The shoulder flexion-extension task (SFE) was similarly executed but with the hand palm facing medially and the thumb pointing forward. One researcher mirrored each task at pace with the participant. Each task was repeated three times but not consecutively as the order of the tasks was randomized.

C. Measurement and processing

Kinematics of the trunk, chest, and right-sided upper extremity was recorded using the Optotrak 3020 system (Northern Digital Inc., Canada) [29]. Single markers were placed on bone landmarks and rigid bodies were placed on soft

Fig. 1. Experimental set up of a FSHD participant about to perform shoulder abduction-adduction (left) and flexion-extension (right).

TABLE I
SINGLE AND RIGID BODY MARKERS

Marker #	Body location
1	Spinal process of 7th cervical vertebra
2	Jugular notch clavicle-sternum
3	Xiphoid process of sternum
4	Acromio-clavicular joint (left)
5	Acromio-clavicular joint (right)
6-8*	Lateral upper arm (right, 1/3 of acromion to lateral epicondy
9	Lateral epicondyle (right)
10	Medial epicondyle (right)
11-13*	Lateral lower arm (right, 1/2 of lateral epicondyle to styloid process of radius)
14	Styloid process of radius (right)
15	Styloid process of ulna (right)
16	Head of the 3rd metacarpal (right)

* Rigid body refers to a rigid cluster of three markers.

tissues on the lateral side of the upper and lower arm as shown in table 1. Humeral elevation was calculated from the recorded kinematics and expressed as joint angle between trunk and humerus where 0° represents the arm straight downward and 180° straight upward.

Surface electromyograms (EMG) of the right side muscles were recorded for the prime humeral elevator/depressors and scapular rotator muscles, i.e. medial Deltoid, Pectoralis Major clavicular head, Latissimus Dorsi, Trapezius Descendens, Trapezius Ascendens, and Serratus Anterior 5-6th rib, and the synergist muscles Biceps Brachii short head and Triceps Brachii long head. Data were captured at 2000Hz using the Delsys Trigno system (Delsys Inc., UK) [30]. Maximum voluntary contractions (MVCs) were recorded beforehand (appendix, table 2). The recorded EMG data were filtered with a 4th order Butterworth 20-450Hz bandpass and a 49-51Hz bandstop filter, rectified, smoothened with a 100ms moving window, normalized to the maximum amplitudes derived from all MVC and task recordings, and filtered with a 4th order Butterworth 5Hz low pass filter. The maximum task-specific muscle activity was extracted as highest normalized amplitude over all task repetitions. Time was normalized to 1001 samples for each repetition ranging from 0 to 100%.

Kinematics and EMG recordings were executed consistently with one researcher placing the markers and electrodes and another research assessing the placement and data quality.

D. Muscle synergy extraction

EMG data were pooled per participant to contain equal samples of both tasks in a single matrix to investigate the shared synergies across humeral elevation planes. Muscle synergies were then extracted using Non-Negative Matrix Factorization (NNMF), which decomposed the matrix into 1 to 8 sets of components consisting of weights and coefficients [24]. These weights and coefficients were converted to a unit vector and represent normalized muscle activity (0-1). Additionally, for each set of components (synergy), the NNMF provided the percentage of variance accounted for of all muscles (VAF) and per individual muscle (VAFM). The minimum required number of synergies per participant were extracted using as thresholds VAF > 90% and VAFM > 75% [24]. The variance accounted for per task was calculated with respect to the reconstructed data (weights * coefficients) for each synergy. Coefficients were then averaged for pooled repetitions per task. Synergies were clustered within each group using an iterative process that matched weights in an ascending order based on Pearson's correlation coefficients.

The muscle synergy extraction procedure was executed for two conditions. One condition included the complete motion and the second condition focused on the upward motion up to 60° humeral elevation.

E. Statistical analysis

Humeral elevation differences between groups were investigated using independent-samples Mann-Whitney U tests. To test the first hypothesis on whether EMG amplitudes of prime movers and synergist muscles would be higher in people with FSHD, the maximum muscle activities were compared using a non-parametric analysis of variance, with Task and Muscle as within-group factors and Group as between-group factor (R v3.5.0, The R Foundation for Statistical Computing, nparLD package) [31]. The Post-hoc tests were performed accordingly between groups using independent-samples Mann-Whitney U tests, and between tasks using related-samples Wilcoxon signed rank tests. Alpha levels were corrected for multiple comparisons and set at 0.025. Effect sizes were expressed as Cohen's d (very small: 0.00-0.01, small: 0.01 - 0.20, medium: 0.20 - 0.50, large: 0.50 - 0.80, very large: 0.80 - 1.20, and huge: >1.20) [32]. Furthermore, the standard error of measurement (SEM) was calculated on the consistency of maximum muscle activity over repetitions for each group and consequently used to calculate standard deviations of mean group differences [33].

To test the second hypothesis on whether muscle synergies were altered or dissimilar in people with FSHD, Pearson's correlation coefficients were used to quantify synergy weight and zero-lag correlation coefficients to quantify synergy coefficient similarities (α: 0.025) [34]. Correlation coefficients values were calculated only for significantly similar synergy weights to minimize type I errors. Additionally, within-group similarity was calculated through the EMG cross-validation method [35], and Pearson correlations for synergy weights only. Differences in within-group similarity from EMG cross-validations were tested with Fisher's least significant difference (LSD) post-hoc test with the number of muscle synergies as a factor (α: 0.025).

III. RESULTS

A. Kinematics

All participants successfully completed all tasks. The control group elevated the humerus significantly higher in SAA to 149±19° (*N=22, Cohen's d:4.28, p<0.001*) and in SFE to 141±17° (*N=22, Cohen's d:3.09, p<0.001*). The FSHD group's maximum humeral elevation was 70±18° and 83±20° during the SAA and SFE task, respectively.

B. Muscle Activity

Maximum muscle activities were significantly different for Task (p<0.010), Muscle (p<0.001), Muscle*Task (p<0.001), Group*Muscle (p<0.001), and Group*Muscle*Task (p<0.001), but not for Group (p:0.248) or Group*Task (p:0.121). Post-hoc tests of the Group*Muscle*Task interaction effect revealed that maximum muscle activities were significantly different between groups (FSHD-control) for Biceps Brachii SFE: +25±2% (*N=22, Cohen's d:1.38 p:0.013*), Trapezius Ascendens SAA: -32±8% (*N=22, Cohen's d:-1.45, p:0.004*) and SFE: -41±6% (*N=22, Cohen's d:-1.95, p:0.001*), and Serratus Anterior SAA: -39±4% (*N=22, Cohen's d:-1.72, p:0.002*) (Fig. 2). Within the control group there was a significant difference between tasks (SAA-SFE) for Trapezius Ascendens: -14±14% (*N=22, Cohen's d:-0.74, p:0.005*) and Latissimus Dorsi: -5±6% (*N=22, Cohen's d:-0.25, p:0.024*). Within the FSHD group significant differences between tasks (SAA-SFE) were found for Biceps Brachii: -12±15% (*N=22,*
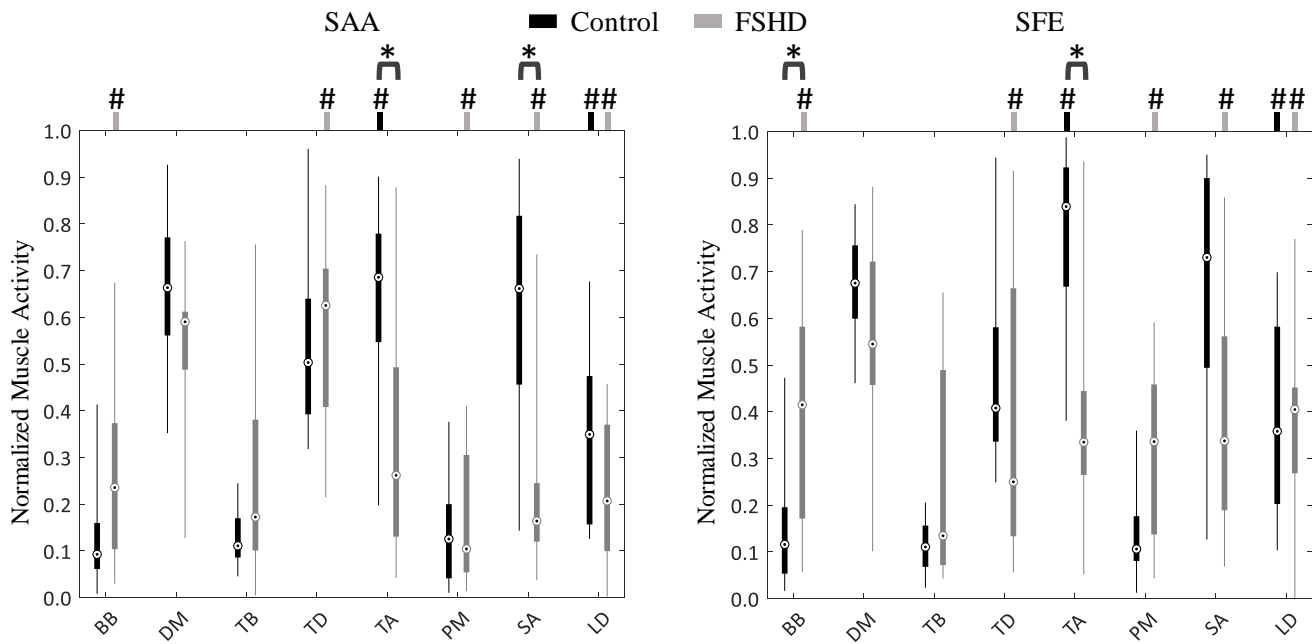
Fig. 2. Boxplots of maximum muscle activity amplitudes of control (black) and FSHD group (grey) for the SAA (left) and SFE (right) tasks (*: significant group differences; #: task differences; p<0.025). Boxes and whiskers indicate minimum, first quartile, median, third quartile, and maximum. BB: Biceps Brachii; DM: medial Deltoid; TB: Triceps Brachii; TD: Trapezius Descendens; TA: Trapezius Ascendens; PM: Pectoralis Major; SA: Serratus Anterior; LD: Latissimus Dorsi.

Cohen's d:-0.55, p:0.010), Trapezius Descendens: +21±25% (*N=22, Cohen's d:0.79, p:0.024*), Pectoralis Major: -13±16% (*N=22, Cohen's d:-0.80, p:0.010*), Serratus Anterior: -19±23% (*N=22, Cohen's d:-0.83, p:0.014*), and Latissimus Dorsi: -17±18% (*N=22, Cohen's d:-0.90, p:0.019*). The SEMs were 1.9% and 3.3% for the control and FSHD group, respectively.

C. Muscle synergies

The number of synergies extracted were equally distributed between the two groups (Fig. 3). In each group at least 90% of the variance was described with one synergy for two participants, two synergies for eight participants, and three synergies for one participant. The control and FSHD group's synergies were clustered into two sets each (Fig. 4, 5). FSHD

Fig. 3. Variance accounted for as means and standard deviation (lines; left y-axis) and number of extracted synergies (bars; right y-axis) of the control (black) and FSHD group (grey). The dashed line indicates the 90% VAF threshold.

participants were also investigated individually and compared to the clustered control synergies (Fig. 4, 5). Appendix Fig. 8 shows the participant-specific synergies.

Synergy #1 on average accounted for 74±19% variance for FSHD participants (controls: 87±9%) in the SAA task and 50±35% VAF (controls: 86±9%) in the SFE task. The VAF per task by synergy #2 was 29±12% for FSHD participants (controls: 15±3%) in the SAA task and 59±27% (controls: 15±6%) in the SFE task. Within-group similarities for synergy weights #1 and #2 were, respectively, for controls R:0.73±0.15 (*N=55*) and R: -0.06±0.37 (*N=36*), and for FSHD R:0.00±0.42 (*N=55*) and 0.08±0.56 (*N=36*). Correlation of synergy weights was not significant for any synergy combination between groups. On an individual level two FSHD participants (#6, #9) showed significant similar synergy weights where synergy #1 correlated with control synergy #2 (*p:0.023, R:0.78 and p:0.001, R:-0.92 for participant #6 and #9, respectively*). Correlation coefficients values for the SAA and SFE tasks were respectively R:0.19 and 0.24 (*FSHD #6, p<0.001 and <0.001*), and R:0.09 and 0.18 (*FSHD #9, p:0.006 and <0.001*).

In the upward motion to 60° humeral elevation condition, at least 90% of the variance was described by two synergies for seven controls and seven FSHD participants, and three synergies for four controls and one FSHD participant. Three FSHD participants did not reach at least 60° in both tasks and were excluded for this condition. Control and FSHD participants' synergies were clustered into three sets each (Fig. 6). Synergy #1 accounted for 63±11% variance for FSHD participants (controls: 62±17%) in the SAA task and 39±10% (controls: 45±16%) in the SFE task. For synergy #2 this was 37±10% and 56±16% (controls: 29±21%, 47±16%) in the SAA and SFE tasks respectively, and 6% and 41% (controls: 24±9%, 21±19%) for synergy #3. Synergy weights showed significant
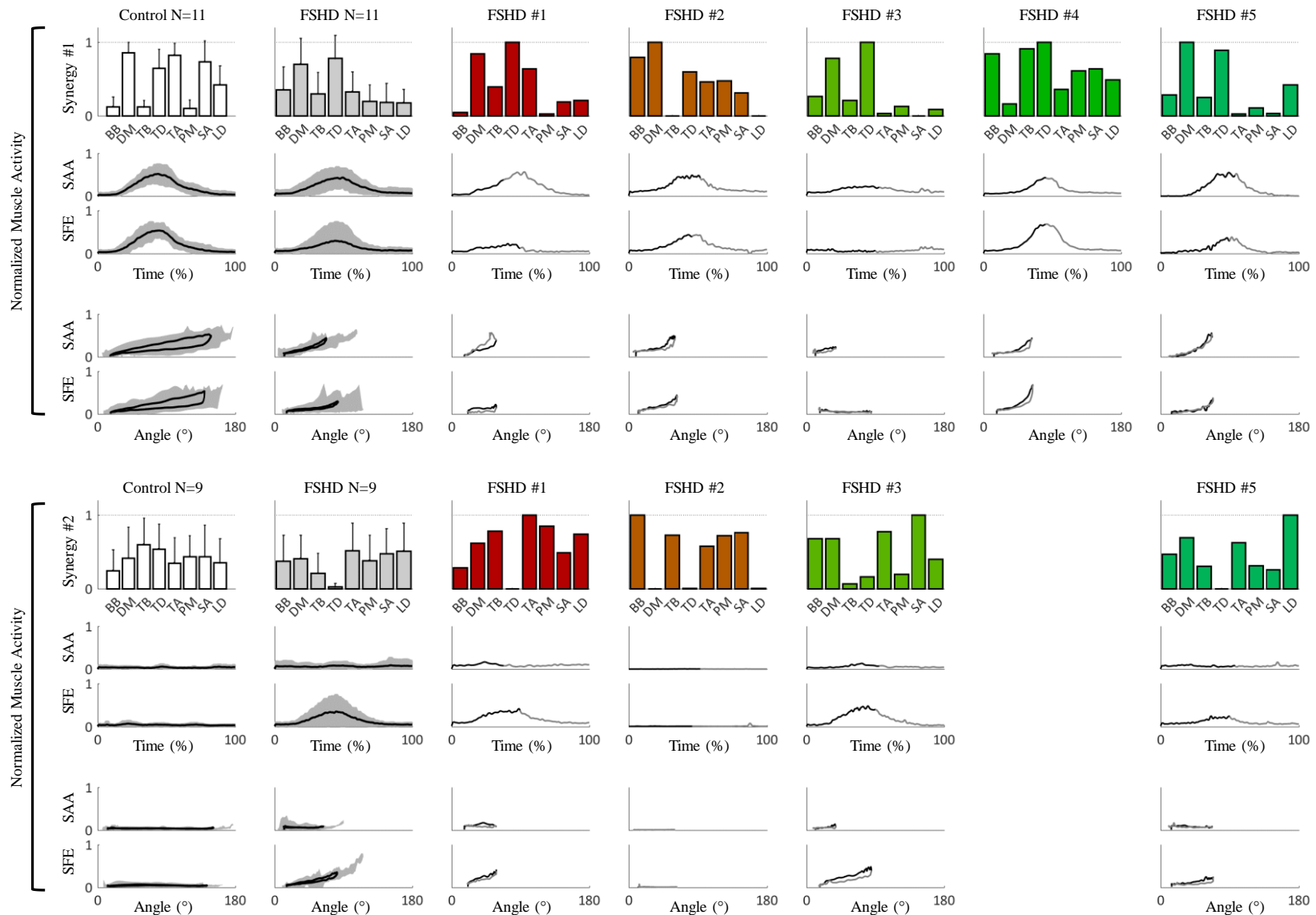
Fig. 4. Muscle synergies no. 1 (top, N=11) and no. 2 (bottom, N=9) of the control group (black) and the FSHD group (grey) and participants 1-5 for the SAA and SFE tasks. The FSHD participants were ranked by averaged humeral elevation in ascending order from left to right (#: participant number). N equals the amount of participants within each clustered synergy. Clustered synergies are presented as mean (rectangles and black thicker line) with standard deviation (bars) or ±95% confidence interval (grey area). Individual synergy coefficients show upward (black line) and downward motion (grey line). Participants #1, 2, 3, and 5 have two synergies, and 4 has one synergy. BB: Biceps Brachii; DM: medial Deltoid; TB: Triceps Brachii; TD: Trapezius Descendens; TA: Trapezius Ascendens; PM: Pectoralis Major; SA: Serratus Anterior; LD: Latissimus Dorsi.

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TNSRE-2018-00556

6

Fig. 5. Continued from previous. Muscle synergies no. 1 (top, N=11) and no. 2 (bottom, N=9) for the remaining FSHD participants 6-11 for the SAA and SFE tasks. The FSHD participants were ranked by averaged humeral elevation in an ascending order from left to right (#: participant number). N equals the amount of participants within each clustered synergy. Clustered synergies are presented as a mean (rectangles and black thicker line) with standard deviations (bars) or ±95% confidence interval (grey area). Individual synergy coefficients show upward (black line) and downward motion (grey line). Participant #6 has three synergies (synergy #3 is presented in appendix fig. 8), 7-10 have two synergies, and 11 has one synergy. BB: Biceps Brachii; DM: medial Deltoid; TB: Triceps Brachii; TD: Trapezius Descendens; TA: Trapezius Ascendens; PM: Pectoralis Major; SA: Serratus Anterior; LD: Latissimus Dorsi.

Fig. 6. Muscle synergies no. 1 (left), no. 2 (middle), and no. 3 (right) of control group (black) and FSHD group (grey) for the SAA and SFE tasks cut up to 60° of humeral elevation. N equals the amount of participants within each clustered synergy. Clustered synergies are presented as a mean (black line) with standard deviation (bars) or ±95% confidence interval (grey area). BB: Biceps Brachii; DM: medial Deltoid; TB: Triceps Brachii; TD: Trapezius Descendens; TA: Trapezius Ascendens; PM: Pectoralis Major; SA: Serratus Anterior; LD: Latissimus Dorsi.
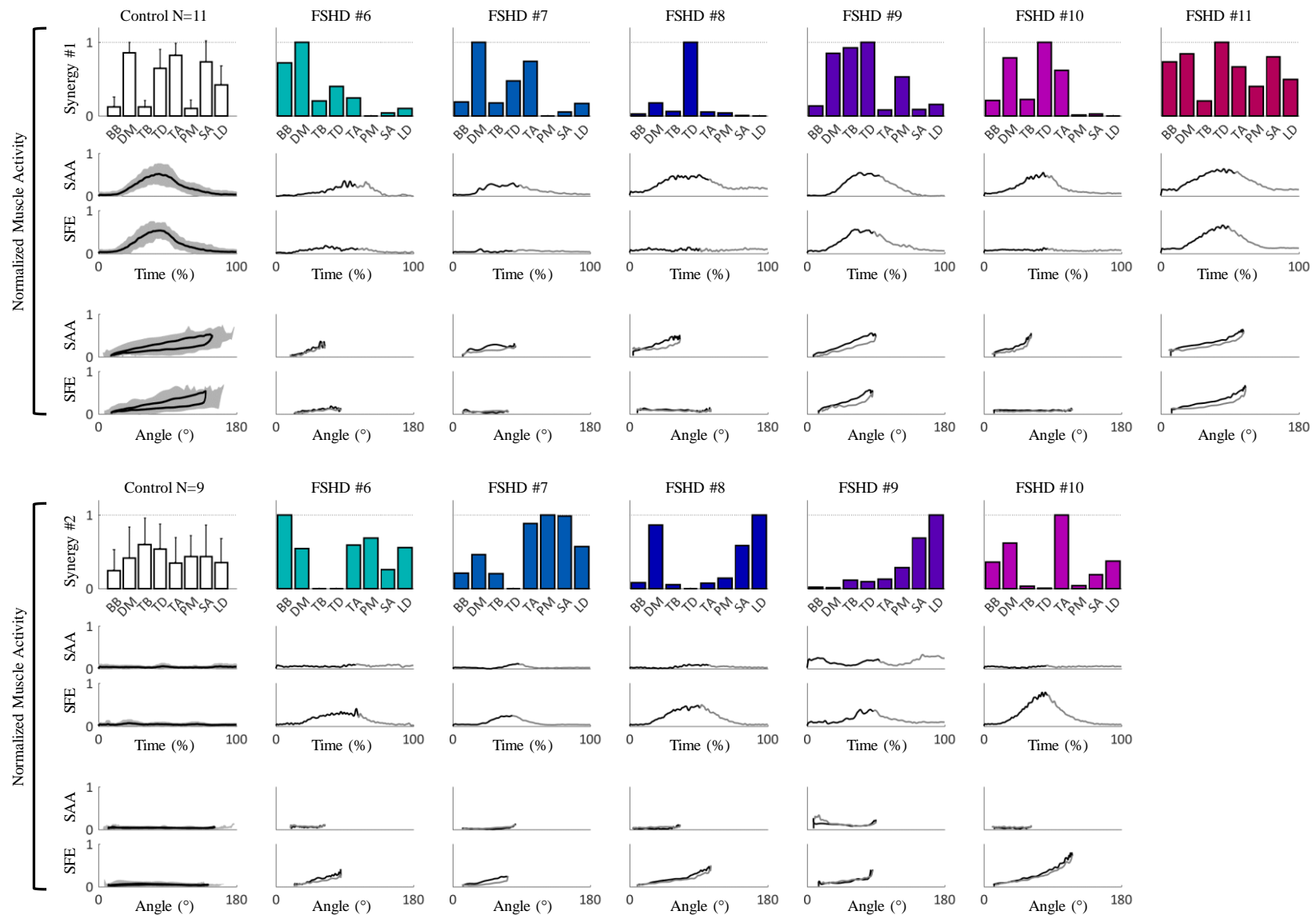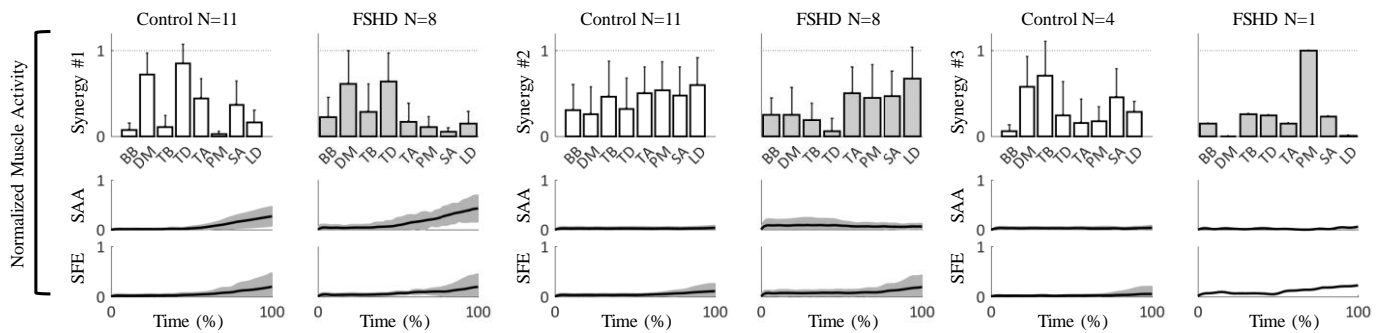
similarities between groups for synergy #1 (*R:0.84, p:0.009*) where correlation coefficients values showed R:0.98 (*p<0.001*) for both tasks.

EMG cross-validations showed that less variance was accounted for by other participants' complete synergy set than one's own set in the control (*p<0.001*) and FSHD group (*p<0.001*, fig. 7). With the exception of controls' synergy #1, other participants' individual synergies accounted for less variance than the complete set (*p<0.001*). Upon further inspection, synergy #2 accounted for an additional 5±2% VAF in controls after which the criteria of >90% was met for 8 participants (Fig. 3). In the upward to 60° humeral elevation condition all factors accounted for less variance than one's own synergy set (*p<0.025*).

IV. DISCUSSION

The activities of eight superficial shoulder muscles were studied to investigate the changes in neuromuscular output in people with FSHD during humeral elevation. We hypothesized that the maximum activities of humeral elevator, scapular rotator and synergist muscles would be increased. This was confirmed for the Biceps Brachii (SFE task only). Contrary to what was expected the activity of the scapular rotators Trapezius Ascendens and Serratus Anterior (SAA task only) significantly decreased in people with FSHD. Additionally, it was hypothesized that the muscle synergies would be altered as a result of the impaired muscle functionality. Along this line, the synergies between groups were found to differ in importance for the complete motion: within group similarity indicated that controls mostly used one synergy for both tasks while the majority of the FSHD group required two task-specific synergies. From the comparable kinematic data and maximum muscle activities it can be concluded that while synergy weights were similar up to 60° humeral elevation, the Trapezius Ascendens and Serratus Anterior contributed on average less to humeral elevation in the FSHD than in the control group. The irregularity of variances accounted for per task by each synergy and the difference in maximum muscle activities, and synergy weights and coefficients, suggest the presence of participant-specific adaptation mechanisms.

The muscle activities of the control group for medial Deltoid (40-69%), Serratus Anterior (60-65%), Trapezius Ascendens

(45-60%), Trapezius Descendens (35-55%), Latissimus Dorsi (10-23%), and Pectoralis Major (5-20%) during shoulder abduction-adduction or flexion-extension were consistent with other literature findings [13], [14], [15]. Maximum elevation angles in the control and FSHD group were also in line with a comparable study [13], while trends in increased activity of Trapezius Descendens and Pectoralis Major found by others [13] were not significant in this study. This could be ascribed to the large variability in muscle activation of people with FSHD [36], and partly to methodological differences in the MVCs protocol used for the Trapezius Descendens. In this study a strap over the shoulder was used to limit the participants' movements during the Trapezius Descendens MVCs recordings, while in [13] the participants' shoulders were manually restrained.

The activities of the lower scapular rotator muscles during humeral elevation tasks in FSHD are presented for the first time
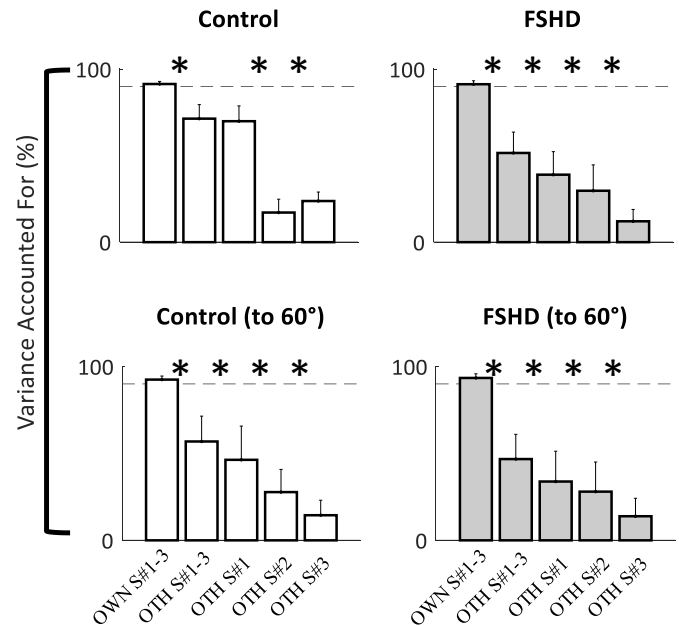
Fig. 7. Variance accounted for as means and standard deviation from EMG cross-validation performed within the control (left) and FSHD group (right) for whole motion (top) and cut to 60° (bottom). Bars show calculations using participants' own synergies (OWN), and from others (OTH) for a complete synergy set (S#1-3) and for individual synergies (S#1, S#2, S#3). Dashed line indicates the 90% VAF threshold. Significant differences were indicated by *.

in this study. The decreased activities of Trapezius Ascendens and Serratus Anterior muscles reveal that these scapular lateral rotators generated a lower force and thus a lower moment to rotate the scapula, a movement which is necessary during humeral elevation [16]. This insufficiency was confirmed by visual observations of very limited scapular rotation in the FSHD group. The decreased activity of these muscles appears to be a characteristic signature of the FSHD disease, which is in contrast with an increased activity of Trapezius Ascendens and Serratus Anterior found in shoulder impingement and post-stroke patients [17], [18], [19], [20], [21], [22]. Ultimately, the inability to laterally rotate the scapula leads to a decrease in humeral elevation. This situation could produce unnecessary stress on the rotator cuff muscles, which provide a stabilizing function of the glenohumeral head and are preserved in FSHD individuals, based on MRI evidence [11], [12]. The increased synergist Biceps Brachii activity likely assisted in the stabilization of the humeral head and the elevation of the humerus within the decreased range of scapular motion [37]. However, a larger variability in muscle contributions did not reveal a clear relationship between the activity of lower scapular rotators or synergist muscles and the amount of humeral elevation.

At the level of intra-task differences between SAA and SFE, a significant increased activity in the FSHD group was found for the Serratus Anterior and Pectoralis Major while an increased activity trend occurred for the Trapezius Ascendens muscle. The higher activity of the Pectoralis Major is consistent with the greater abduction moment required during forward flexion. Furthermore, more scapulothoracic internal rotation is known to occur in healthy shoulders during shoulder flexion-extension than abduction-adduction [16], while external rotation of the scapula increases following Serratus Anterior fatigue [38]. A higher activity of the Trapezius Ascendens and Serratus Anterior during shoulder flexion-extension is therefore consistent with the requirements for more internal scapula rotation and joint stability.

In order to understand whether the coordinated activity, i.e. synergy weights, of selected muscles underlines possible compensatory strategies in the FSHD group, a muscle synergy analysis was carried out and presented here for the first time in this population. The synergies accounting for the highest proportion of the VAF (Fig. 4, 5) showed a changed coordinating action of humeral elevator and scapular rotator muscles. Specifically, synergy #1 for the control group was most likely responsible for glenohumeral elevation, scapula rotation and scapula stabilization, as exemplified by the main contributions of the Deltoid Medial, Trapezius Descendens and Ascendens, Serratus Anterior, and Latissimus Dorsi muscles. Synergy #1 for the FSHD group showed involvement of the Deltoid Medial and Trapezius Descendens and was therefore most likely responsible for glenohumeral elevation and scapula upward rotation. Contributions from the Trapezius Ascendens, Serratus Anterior, and Latissimus Dorsi muscles appeared diminished compared to the control group, reflecting the differences found in maximum muscle activity. The controls' second synergy was characterized by low muscle activation and

follows from the methodological choice of accounting for >90% variance of all muscles. We postulate that this second synergy is a collection of short activation bursts (<20%) from different muscles, possibly to stabilize or facilitate the movement. Eight out of eleven FSHD participants used a second synergy with distinct coefficients for the SFE task. This second synergy was most likely responsible for scapula rotation and stabilization. This synergy also differs from the first in the contributions from Trapezius Ascendens and Serratus Anterior, reflecting the task-specific differences found in maximum muscle activity. Additionally, FSHD participants who applied this second synergy had higher humeral elevation angles. Similarly to what was found for the maximum activity, no clear relationships was present between the humeral elevation angles achieved and the amount of required synergies and/or involvement of scapula rotator/stabilizer muscles. The variety in muscle synergies compositions shows evidence that muscle control is less consistent in FSHD.

The synergy coefficients of similar synergy weights correlated poorly between groups. However, considering that the similarities were computed between two synergies of a high VAF proportion (FSHD) vs. a clustered synergy of a low VAF proportion (control), it is questionable whether the comparison is representative of the change at a group level. Additionally, EMG cross-validations indicated a larger data similarity within the controls than the FSHD group, revealing a higher consistency in muscle activation in the former group. Future analysis should specifically focus on evaluation of within-group similarity of synergy weights and coefficients. In summary, coordination differences in FSHD appear to reflect the physiological changes of muscles due to the disease.

On the basis of the above findings it is evident that FSHD can lead to alterations in the coordination of muscle groups and lead to altered function and thus performance in some individuals. Muscle strengthening therapy, including scapular control, is sometimes considered part of the rehabilitation treatment for impingement and scapular winging [42], [43], [44]. Although scapular control therapy remains a debated topic [45], future research should explore whether this therapy could be effective in people with FSHD [36], [46]. Given the limited function of the scapular rotators it is likely that therapeutic decisions should be made on an individual level, after careful assessment of the muscles' coordination using a methodology similar to the one proposed in this study.

A. Limitations

Muscle synergy analysis was shown to be sensitive to detect changes in motor output with respect to internal/external factors, however the technique has its limitations. The outcomes can be influenced by the choices made in EMG processing, NNMF settings, and threshold of VAFs [39], [40], [47], [48]. For example, a lower VAF threshold would reduce the required number of synergies, possibly oversimplifying the motor output. To overcome this problem, this study uses two thresholds to ensure the variance of all muscles have been accounted for on a collective as well as a singular level [24]. In addition, the statistical approach was thorough and ensured that

the limitations did not affect the conclusions.

The number of muscle synergies were inconsistent between participants and resulted in two clustered synergies of eleven and nine participants. However, this can be explained by individual characteristics, unrelated to disease effects [39], [40]. Furthermore, the total number of synergies were equal between the groups. Nonetheless, this could have resulted in the large within-group variances, specifically in muscle synergy weights, where a common coordinating activity is only evident for selected muscles [41]. The presented clustering method is suitable for simple movements as examined in this study, but arguably not when multiple synergies are needed, for example during more complex motions. Other cluster analysis methods can be used to pool synergies based on more distinct weights [27], [28] and are recommended in future research.

V. Conclusion

People with FSHD showed motor output alterations during humeral elevation, which were often movement- and participant-dependent. In general, the lower scapula rotators showed decreases in activity, with compensatory increase of a synergistic upper arm muscle. A group*muscle*task interaction effect was accompanied with increased activities of the lower scapula rotators, and synergistic chest and upper arm muscles during shoulder flexion-extension compared with abduction-adduction. The large group variances indicate that individual characteristics have a large influence on motor output. An assessment of the muscles' coordination is recommended to reveal individual synergies and to design evidence-based therapy for the management of the condition.
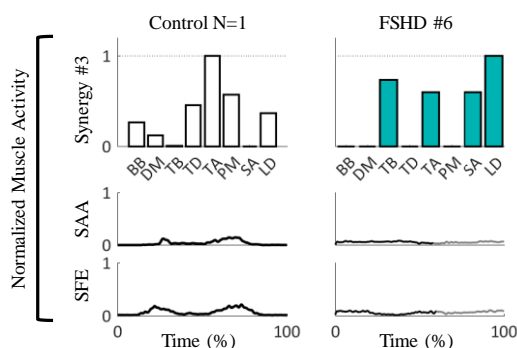
Appendix

Fig. 8. Participant-specific muscle synergy of a control participant (black, 9% VAF) and FSHD participant #6 (dark cyan, 19% VAF) for the SAA and SFE tasks. N equals the amount of participants within the clustered synergy. Individual synergy coefficients show upward (black line) and downward motion (grey line). Participant #6 has three synergies. BB: Biceps Brachii; DM: medial Deltoid; TB: Triceps Brachii; TD: Trapezius Descendens; TA: Trapezius Ascendens; PM: Pectoralis Major; SA: Serratus Anterior; LD: Latissimus Dorsi.

TABLE II
MAXIMUM VOLUNTARY CONTRACTION PROTOCOL

Muscle	Instructions
Biceps Brachii short head (BB)	Position: Upper arm is alongside the torso. Elbow is flexed at 90° and forearm temporarily supported by a researcher. A strap on the wrist prevents elbow flexion. Execution: Flex the elbow against the strap.
Medial Deltoid (DM)	Position: Upper arm is abducted at 60°. Elbow is flexed at 90° with hand palm downwards. A strap on the upper arm prevents abduction. Execution: Abduct the arm against the strap.
Triceps Brachii long head (TB)	Position: Upper arm is abducted at 90°. Elbow is flexed at 90° with hand palm downwards. Execution: Extend the forearm against the resistance provided by a researcher.
Trapezius Descendens (TD)	Position: Upper arm is alongside the torso. Elbow is fully extended. A strap is placed above the shoulder and medial to the acromion. Execution: Pull the shoulder upwards against the strap.
Trapezius Ascendens (TA)	Position: Both upper arms are alongside the torso. Elbows are fully extended. Execution: Bend the trunk slightly forward and elevate the arms to form a straight line from the fingertips to the hips.
Pectoralis Major clavicular head (PM)	Position: Upper arm is abducted at 90°. Elbow is flexed at 90° with hand palm downwards. Execution: Adduct the arm to the sagittal plane against the resistance provided by a researcher.
Serratus Anterior 5-6th rib (SA)	Position: Upper arm is alongside the torso. Elbow is fully flexed. Execution: Pull the shoulder down by pushing the elbow towards the hip against the resistance provided by a researcher.
Latissimus Dorsi (LD)	Position: Upper arm is abducted at 90°. Elbow is flexed at 90° with hand palm downwards. Execution: Adduct the upper arm against the resistance provided by a researcher.

MVCs were executed while seated, with two repetitions and 2 minutes rest between repetitions.

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