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Biomechanical analysis of upper limb of patients with Duchenne muscular dystrophy in chosen functional tasks: A multi-case study

Agnieszka Sobierajska-Rek [a], Wiktoria Wojnicz [b], Bartłomiej Zagrodny [c], Michał Ludwicki [c,*], Katarzyna Pytka [b], Joanna Jabłońska-Brudło [a]

[a] Department of Rehabilitation Medicine, Faculty of Health Sciences with Institute of Maritime and Tropical Medicine, Medical University of Gdansk, Gdansk, Poland
[b] Faculty of Mechanical Engineering and Ship Technology, Gdansk University of Technology, Gdansk, Poland
[c] Mechanical Engineering Faculty, Lodz University of Technology, Lodz, Poland

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ABSTRACT

Background: The main symptom of Duchenne muscular dystrophy is progressive wasting of muscle tissue. Current research on motor measurements for these patients lacks assessments of the whole trajectories of the functional motions over time in clinical conditions. The main goal of this study was to compare the upper limb movement strategy of patients with Duchenne muscular dystrophy and a healthy Control by assessing the contributions of chosen joint angles and contributions of muscles activity in the functional motions of activities of daily living.
Method: A novel score of kinematic and electromyographic patterns is proposed, and results are assessed for a whole trajectory of tested upper limb for five patients and a healthy Control. Contributions of four joint angles and four surface muscle activities are assessed for two relations – kinematic control (a relation between muscle activity and displacement of the wrist) and dynamic control (a relation between muscle activity and displacement of the center of mass of the upper limb).
Findings: In vertical motions, higher mobility in the shoulder and elbow was observed in patients with lower Brooke scores. In motions involving horizontal movement of a weight, the contribution of the elbow and the trapezius were higher in all patients vs the Control.
Interpretation: The proposed method revealed significant differences in muscle activity and upper limb movement patterns in patients with Duchenne muscular dystrophy compared with a healthy Control. Results of the assessment of contributions in kinematic control and dynamic control show individual movement strategies of patients with Duchenne muscular dystrophy.

1. Introduction

Duchenne muscular dystrophy (DMD) is one of the rare neuromuscular diseases and the most common form of muscular dystrophy in male children with an incidence of 1 in 3500–6000 live births (Alison et al., 2012; Bendixen et al., 2016; Mendell et al., 2012). It is an X-linked recessive disorder characterized by progressive muscle wasting and weakness (Duan et al., 2021; Hiebeler et al., 2023; Rinaldi et al., 2020; Yoon et al., 2022). Despite some therapies being in clinical development (Duan et al., 2021; Mercuri et al., 2019) there is currently no cure for DMD, and treatment is mainly aimed at delaying disease progression and preserving gait (De Souza et al., 2016), and all functional abilities (Attias et al., 2016; Bendixen et al., 2016; Case et al., 2018; Eagle et al., 2007; Yoon et al., 2022). The first symptoms of the disease usually appear between the ages of 2 and 6 years (Hiebeler et al., 2023), and loss of independent ambulation is observed in 10–14 year-old males. As the disease progresses, patients with DMD gradually lose muscle strength in the upper extremities (UE), which can lead to an inability to reach overhead and self-feed (hand-to-mouth movement) (Sobierajska-Rek et al., 2024). Difficulties with reaching objects at a waist level are also observed (Birnkrant et al., 2018; Koeks et al., 2017; Mayhew et al., 2013). This is mainly caused by muscle weakness in the elbow joint, as well as increasing contractures developing in elbow and wrist joints, and long finger flexors. In the late non-ambulatory stage patients with DMD may struggle with grasping or pushing buttons, as well as experiencing gastrointestinal problems, swallowing disorders, heart failure, and respiratory insufficiency (Connolly et al., 2015; Maria et al., 2012). Moreover, a questionnaire from the International Classification of

* Corresponding author.
E-mail address: michal.ludwicki@p.lodz.pl (M. Ludwicki).

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Functioning, Disability, and Health (ICF) provides insight into the changing functions of the upper limb during DMD at three different levels: body functions and structures, activities, and social participation (Janssen et al., 2014). Results of this questionnaire manifest that pain, stiffness, and activity limitations in the UE increase with disease progression. Also, the range of motion (ROM) of the UE of each patient with DMD is limited in comparison to the ROM of healthy subjects (Khallaf et al., 2017; Pangalila et al., 2011).

It should be emphasized that DMD is a progressive disease. That is why the motion performances of each patient with DMD should be monitored while performing activities of daily living (ADL). Moreover, to establish the proper rehabilitation strategy a detailed analysis of upper limb kinematics should be done to identify a disease progression (Janssen et al., 2017; Maletsky et al., 2007; Romilly et al., 1994; Rosen et al., 2005; Šenk and Chèze, 2010; Spörri et al., 2016; Summers et al., 2008; van der Kruk and Reijne, 2018). Nowadays, to assess the mobility of the upper limb of a patient with DMD clinicians use clinical scales and functional tests, e.g., the Vignos (Maria et al., 2012), Brooke (Mercuri et al., 2012), or PUL scale (Mayhew et al., 2020; Sobierajska-Rek et al., 2024). However, the results and repeatability of these examinations are mainly dependent on the experience of the clinician conducting assessments. Moreover, these results do not inform about inter-joint coordination changes and compensatory movement changes (Carpinella et al., 2020; Naarding et al., 2022; Ricotti et al., 2023). To date, there are only a few studies that share the results of conducting clinical assessments on patients with DMD with the use of a precise engineering measurement. The analysis of these results provides more reliable and unbiased data along with minimalization of subjective assessments (Carpinella et al., 2020; Mercuri and Mazzone, 2011; Naarding et al., 2022; Ricotti et al., 2023; Wojnicz et al., 2022). However, the literature review lacks reports describing the results of the assessment of the whole trajectories of tested functional motions over time in clinical conditions. It is worth paying attention that patients with muscle atrophy and other types of dystrophies, that cause a decrease in muscle strength and ROM, cannot be treated as a coherent group in terms of kinematic or activity parameters (based on electromyography (EMG) measurement) and especially they may vary significantly regarding weight, height and other anthropometric parameters. Because of corticosteroid treatment and low physical activity, many of them demonstrate overweight or obesity. Furthermore, in some cases in the late stage of the disease, difficulties with eating and severe scoliosis may lead to malnutrition and underweight (Wernio et al., 2024). Due to the constant corticosteroid intake patients may also present growth deficiency and pubertal delay (Wood et al., 2016). The clinical course of the disease varies significantly among patients and is dependent on dystrophin mutations, treatment with corticosteroids or other disease-modifying agents, and variations in clinical practices (Szabo et al., 2021). This poses a challenge for researchers in collecting a larger group of volunteers with a similar level of disease progression and comparable anthropometric indexes, as well as in performing multiple repetitions of tested movements (Carpinella et al., 2020; Germanotta et al., 2023; Pauk et al., 2023).

The main motivation of this study was to create a tool that may allow clinicians to receive information about changes in the inter-joint coordination and compensatory movement of patients with DMD. In this study, we propose a novel scoring of kinematic and surface electromyography patterns allowing to assess contributions (participations) of four joint angles of an upper limb and four chosen superficial muscle activities in each tested performance by considering the whole track of each tested ADL motion.

2. Methods

The scope of this study involved examining five non-ambulant patients with DMD (15.2 ± 2.2 years, 73.60 ± 22.06 kg, 162.20 ± 3.49 cm) that were scored by using Vignos and Brooke scales. All selected volunteers were males with diagnosed DMD, confirmed by genetic testing and/or muscle biopsy, and without difficulties with cooperation. The control group was composed of 12 healthy teenager/adolescent boys with a lack of postural disorders (14.3 ± 1.8 years, 66.87 ± 21.5 kg, 170.45 ± 10.66 cm). All volunteers (control group and patients with DMD and their parents) provided written informed consent following procedures approved by the agreement of the Ethic Committee of Medical University of Gdansk NKBBN/23/2019, NKBBN/23–708/2019, NKBBN/23–409/2020.

To compare the results of the patients with DMD, who were in different stages of disease progression, the clinicians chose from the control group the one control subject (healthy Control), which had a right-limb dominance along with an anthropometric proportion of the body that was similar to the tested patients with DMD. Anthropometric data with Vignos (ambulatory status) and Brooke scale results (upper limb function) of tested patients with DMD and healthy Control are presented in Table 1.

In this study we tested three types of motion: 1) functional vertical motions (V1 - lifting the weight of 50 g from the waist level to the shoulder level; V2 - lifting the weight of 200 g from the waist level to the shoulder level); 2) functional horizontal motions at waist level (H1 - moving the weight of 100 g on the table; H2 - moving the weight of 200 g on the table; H3 - moving the weight of 500 g on the table; H4 - moving the weight of 1000 g on the table; 3) functional complex motions (V3 - raising the hand to the mouth with a weight of 50 g from the waist level; H5 - follow the track by a finger at a waist level). During testing each patient used their own wheelchair without armrests and each control subject sat on an adjusted stool without a backrest. Each tested person sat at the table adjusted to be even with his waist.

Considering recommendations given for healthy populations and patients with neuromuscular disorders in (Alt Murphy et al., 2011; Centen et al., 2017; Germanotta et al., 2023; The FDG Robotic Rehabilitation Group et al., 2020), the considered number of corrected trials performed by a Control equaled five, and each patient with DMD equaled three. This number of trials allowed for minimizing fatigue and avoiding the effect of learning. Results of the motions performed by the tested subject and accepted by the clinicians are given in Table 2. It is worth emphasizing that recorded motions were spontaneous ones performed by Control and each tested patient after the verbal instruction was given by a medical doctor without any preliminary training (recorded motions were results of own perception of each tested subject). During the execution of the motion, a medical doctor did not give any suggestions on how to initiate and correct the motion performance.

To record kinematic data of the upper body part of each tested subject we used the OptiTrack Flex 13 system (6 cameras, 120 Hz) and dedicated software *"OptiTrack Motive:Body"* (NaturalPoint, Inc. P.O. Box 2317, Corvallis, OR 97339). It is worth emphasizing that this system can be used as a mobile set (in clinics) or a stationary one (in laboratories). To identify segments of each arm and trunk a marker protocol composed of 20 markers was used (according to (Alt Murphy et al., 2011)). To analyze recorded kinematic data, we processed these data by using the 4th-order Butterworth filter with a 5 Hz cut-off frequency (Robertson et al., 2013; Winter, 2009; Yu et al., 1999). In this study, we analyzed the kinematic trajectory of ADL motions in patients with DMD being in different stages of disease progression by focusing on four tested joint angles of the right upper limb – elbow joint: flexion-extension

Table 1
Anthropometric data and Brooke/Vignos scale scores.

	Brooke/Vignos [–]	Arm length [cm]	Forearm length [cm]
Control	–/–	32	27
DMD1	3/9	32	26
DMD2	4/9	31	24
DMD3	5/9	30	25
DMD4	5/9	31	22
DMD5	5/9	35	28

Table 2
Motion examined in the study (lack of symbol "+" means that the tested patient with DMD could not perform motion in a correct way due to pain and contractures).

	V1	V2	V3	H1	H2	H3	H4	H5
Control	+	+	+	+	+	+	+	+
DMD1	+	+	+	+	+	+	+	+
DMD2	+	+	+	+	+	+	+	+
DMD3	+	+	+	+	+	+	+	+
DMD4				+	+	+	+	+
DMD5					+	+	+	+

(EFE); shoulder joint: rotation (SRot), flexion-extension (SFE), and abduction-adduction (SAA).

EMG data of four chosen superficial muscles (trapezius (EMG1), lateral triceps brachii (EMG2), anterior deltoid (EMG3), and biceps brachii (EMG4)) had been recorded using the Noraxon MyoTrace400 system (designed in accordance with IEC60601–2-40) and commercial software "*Noraxon MyoResearch XP Clinical Edition*" (Noraxon, Scottsdale, AZ, USA). This wiring system synchronically collected data from four channels with a 1000 Hz sampling frequency. Each channel had an anti-aliasing filter working in [10;500] Hz frequency range. Specifications of each channel preamplifier were the following: common mode rejection ratio (CMRR) exceeded 100 dB, input impedance was greater than 100 MΩ, baseline noise was less than 1 μV RMS, base gain was 500 and input range was ±3.5 mV (Troka et al., 2022). The analog-to-digital conversion of each channel had a 16-bit resolution. To collect EMG data two types of disposable self-adhesive Ag/AgCl snap-on electrodes with electrolytic gel were used: 1) Noraxon Dual Electrode (1 cm diameter of circular wet gel conductive; 2 cm inter-electrode distance; 4 cm × 2.2 cm adhesive area); 2) Noraxon Single Electrode (1 cm diameter of the circular wet gel conductive area, 3.9 cm diameter of circular adhesive area; this electrode was used as a reference one). The reference electrode was attached to the medial clavicular head or olecranon. Dual electrodes were placed over tested muscle bellies on the properly prepared skin according to SENIAM recommendations (Merletti and Cerone, 2020; Stegeman and Hermens, 2007). The medical tape was used to secure electrodes and cables to reduce motion artifacts.

The MyoResearch XP software was used to collect and record raw EMG data, and to process these data by applying rectification and smoothing (Root Mean Square (RMS) algorithm with a 50 ms non-overlapping window). It is worth noting that each EMG channel direct component (DC), which corresponded to background noise, was cut back from the raw EMG data collected over testing before signal processing (rectification and smoothing). Using the "zero offset" function of MyoResearch XP software, this DC component was determined from EMG data collected over a preliminary time interval, in which all tested muscles were in a relaxation state (Troka et al., 2022).

The MATLAB software was used to obtain normalized processed EMG data by implementing own scripts and considering electromechanical delay equaling 50 msec. These scripts were created to perform the following steps: 1) to normalize the time scale of processed EMG data to the timing of motion (percentage of motion) (Wojnicz et al., 2022); 2) to normalize the amplitude of processed EMG data; 3) to take into account the EMG onset (threshold) and to choose significant muscle activity. Considering the fact that during one visit each examined patient with DMD was not able to simultaneously perform tested ADL motions and maximum voluntary contraction (MVC) without fatigue, the processed EMG data were normalized with respect to the maximum value registered during the whole test (reference voluntary contraction) (Barański et al., 2024; Ellis et al., 2017; Wojnicz et al., 2022). Also, the EMG onset equaled 0.005 of RMS had been implemented according to (Ellis et al., 2017; Glickman et al., 2020; Rashid et al., 2019; Stegeman and Hermens, 2007; Voet et al., 2022), and in the next step, two clinicians used visual inspection to choose significant muscle activity from all processed EMG data (RMS EMG) along the guidelines given in (Glickman et al., 2020; Voet et al., 2022). This chosen muscle activity (presented in Supplement in section S.4) had been used to calculate an EMG index (described below).

A novel scoring of kinematic and EMG patterns proposed in this paper is based on the hypothesis that a tested subject (a patient with DMD, healthy Control) performs ADL functional motions by controlling a displacement of the tested upper limb wrist joint (WJ) and/or a displacement of the center of mass (COM) of this limb through (Fig. 1A): a) configurations of upper limb joint angles involved in the performance of the tested motion (EFE, SRot, SFE, and SAA) (called **a relation A**); b) activation of muscles acting on the upper limb joints involved in the performance of the tested motion (EMG1, EMG2, EMG3, and EMG4) (called **a relation B**). Results of both relations are presented as separate contributions (participations) of four joint angles (results of a relation A) and separate contributions of four surface muscle activity (results of a relation B). All contributions are estimated by applying a linear piecewise multi-regression calculated in each tested window composed of 10 frames (this length of window equals 1 % length of each tested motion track) according to (Wojnicz et al., 2022). In relation A and relation B independent variables are: 1) four joint angles (EFE, SRot, SFE, and SAA) (in relation A); 2) four muscle activities (EMG1, EMG2, EMG3, and EMG4) (in relation B). In both relations, the displacement of the tested upper limb wrist joint (WJ) and the displacement of the center of mass (COM) of this limb were treated as dependent variables.

A novel scoring is composed of three kinematic indexes (1,2,3) and one EMG index (4):

1) ***The kinematic synergy of two joint angles over time*** is associated with joint coordination (this synergy is assessed based on cross-correlation results of each pair of considered joint angles).
2) ***The accumulated joint synergy of four joint angles over time*** **(relation A)** is assessed from the linear piecewise multi-regression and estimates the contributions of the tested joint angles in examined functional motions by determining ***joint-kinematic relations*** (statistically significant correlations between tested joint angles and WJ displacement) and ***joint-dynamic relations*** (statistically significant correlations between tested joint angles and COM displacement).
3) ***The predominant type of control of accumulated joint synergy of four joint angles over time*** reveals whether the tested subject had predominant joint-kinematic relations or predominant joint-dynamic relations while performing functional tests (predominant type of control is assessed based on average results of statistically significant coefficients of determination R^2 referring to the accumulated joint synergy of four joint angles over time).

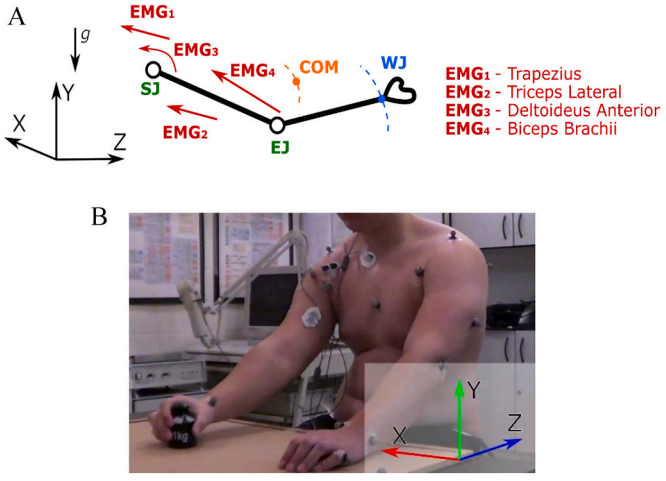

Fig. 1. (A). Schematic visualization of the examined upper limb. (B). Axis directions of the chosen subject tested in this study.

4) ***The accumulated EMG synergy of four superficial muscles over time* (relation B)** is assessed from the linear piecewise multi-regression and estimates the contributions of tested muscles in examined functional motions by determining ***EMG-kinematic relations*** (statistically significant correlation between the activity of tested muscles and WJ displacement) and ***EMG-dynamic relations*** (statistically significant correlation between the activity of tested muscles and COM displacement).

Additionally, in the Supplement, we give a description of active functional ROM test results (assessed from the active motions of each tested subject) (Supplement, section S.1).

To estimate ***three kinematic indexes*** of proposed novel scoring the normalized kinematic data were used. These data were calculated for each trial in two steps by proving that minimum displacement equaled zero: first, displacements were divided by the maximum value of displacement that occurred in a considered trial; next, these data were normalized to the time scale, i.e. percentage of motion. In each trial, a displacement of the wrist joint (WJ) was recorded in the Cartesian coordinate system XYZ (Fig. 1B). The displacement of COM related to the X, Y, and Z axes was estimated based on the method of body segmentation. In the scope of this study, the following normalized kinematic data were analyzed: 1) the *x*-th, *y*-th, and *z*-th displacement of the wrist joint (WJx, WJy, WJz); 2) the *x*-th, *y*-th, and *z*-th displacement of the upper limb COM (COM_x, COM_y, COM_z).

The kinematic synergy of two joint angles over time was assessed from processed kinematic data by using a cross-correlation function implemented in MATLAB scripts. A result of a kinematic synergy presents a measure of matching of two signals in a time series. This measure presents as a coefficient of cross-correlation (CC), and its value is used to identify whether there is similarity (kinematic synergy) in time between two tested joint angles and is interpreted as a coordination index (Carpinella et al., 2020).

To estimate ***the accumulated joint synergy of four joint angles over time* (relation A)** and to determine ***joint-kinematic relations*** (whether tested joint angles are correlated with the displacement of the wrist joint) along with ***joint-dynamic relations*** (whether the joint angles are correlated with the displacement of COM of the limb), the sets of dependent variables $\boldsymbol{y}$ were composed of displacements of the wrist joint (WJx, WJy, WJz) and COM of the right upper limb (COMx, COMy, COMz). A set of independent variables was composed of four joint angles (EFE, SRot, SFE, SAA). The following piecewise multi-regression relationship was used to assess the contributions of ***a joint synergy*** for each set of dependent variables $\boldsymbol{y}$ in each tested window:

$$\begin{gathered}\boldsymbol{y} = a_0 + (a_1 \bullet \mathrm{EFE}) + (a_2 \bullet \mathrm{SRot}) + (a_3 \bullet \mathrm{SFE}) + (a_4 \bullet \mathrm{SAA}) = \\ = y_0 + y_1 + y_2 + y_3 + y_4, \\ a_1 \bullet \mathrm{EFE} = y_1, \\ a_2 \bullet \mathrm{SRot} = y_2, \\ a_3 \bullet \mathrm{SFE} = y_3, \\ a_4 \bullet \mathrm{SAA} = y_4,\end{gathered} \tag{1}$$

where: y_j (for $j = 0, 1, 2, 3, 4$) describes the j-th participation in the tested dependent variable $\boldsymbol{y}$; a_0 implies the part describing the contribution of the motion of the trunk, non-monitored joints of upper limb and supination-pronation of the forearm ($a_0 = y_0$); a_i ($i = 1, \ldots, 4$) describes the coefficient of the i-th joint angle that defines participation of this angle in the tested motion.

In the next step, considering results obtained from Eq. (1), we took into account only statistically significant results with coefficients of determination $R^2 \geq 0.75$. These results were used to calculate separate percentage participations of each chosen joint angle y_j with respect to the X, Y, and Z coordinate in each tested window:

$$part_{y_{i\_WJ_{dim}}} = \frac{y_{i\_WJ_{dim}}}{\sum y_{i\_WJ_{dim}}} \bullet 100\%, \tag{2}$$

$$part_{y_{i\_COM_{dim}}} = \frac{y_{i\_COM_{dim}}}{\sum y_{i\_COM_{dim}}} \bullet 100\%, \tag{3}$$

where dim denotes *x*-th, *y*-th, and *z*-th components.

Next, we used results obtained from the Eq. (2) to estimate the value of ***joint-kinematic relations of accumulated joint synergy for each joint angle over time***:

$$accumulated_{y_{i\_kinematic}} = \frac{1}{3} \bullet \left(\left| part_{y_{i\_WJx}} \right| + \left| part_{y_{i\_WJy}} \right| + \left| part_{y_{i\_WJz}} \right| \right), \tag{4}$$

and we used results obtained from the Eq. (3) to assess the value of ***joint-dynamic relations of accumulated joint synergy for each joint angle over time***:

$$accumulated_{y_{i\_dynamic}} = \frac{1}{3} \bullet \left(\left| part_{y_{i\_COMx}} \right| + \left| part_{y_{i\_WJy}} \right| + \left| part_{y_{i\_WJz}} \right| \right). \tag{5}$$

The predominant type of control of accumulated joint synergy of four joint angles over time for each tested subject and each tested motion is assessed by taking into account only statistically significant results of the accumulated kinematic synergy of four joint angles over time. Each predominant type of control was calculated as an average of statistically significant coefficients of determination R^2 (i.e., $R^2 \geq 0.75$) estimated separately for each coordinate (X, Y, Z) in each kinematic relation (R2WX, R2WY, R2WZ) and each dynamic relation (R2CMX, R2CMY, R2CMZ).

To assess ***the accumulated EMG synergy of four superficial muscles over time* (relation B)** and to determine ***EMG-kinematic relations*** (whether four muscle activations are correlated with the displacement of the wrist joint) along with ***EMG-dynamic relations*** (whether four muscle activations are correlated with the displacement of COM of the tested limb) the normalized processed EMG data were used. Contributions of an ***EMG synergy*** for each set of dependent variables $\boldsymbol{y}$ (described in Eq. (1)) were assessed from the piecewise multi-regression relationship in each tested window by using the following relationship:

$$\begin{gathered}\boldsymbol{y} = a_{0\mathrm{EMG}} + a_{1\mathrm{EMG}} \bullet \mathrm{EMG}_1 + a_{2\mathrm{EMG}} \bullet \mathrm{EMG}_2 + a_{3\mathrm{EMG}} \bullet \mathrm{EMG}_3 + a_{4\mathrm{EMG}} \bullet \mathrm{EMG}_4 = \\ = y_{0\mathrm{EMG}} + y_{1\mathrm{EMG}} + y_{2\mathrm{EMG}} + y_{3\mathrm{EMG}} + y_{4\mathrm{EMG}}\end{gathered} \tag{6}$$

where: y_{jEMG} (for $j = 0,1,2,3,4$) describes the j-th participation (contribution) in the value of the tested kinematic data $\boldsymbol{y}$; $a_{0\mathrm{EMG}}$ implies the part describing the contribution of the factors that are not related with the tested muscles ($a_{0\mathrm{EMG}} = y_{0\mathrm{EMG}}$); $a_{i\mathrm{EMG}}$ ($i = 1,..,4$) describes the coefficient of the i-th muscle activation $\mathrm{EMG_i}$ (this coefficient depends on this muscle lever arm). The product between this coefficient $a_{i\mathrm{EMG}}$ and muscle activation EMG_i defines a contribution of this i-th muscle in the motion performance, i.e. $a_{1\mathrm{EMG}} \bullet \mathrm{EMG}_1 = y_{1\mathrm{EMG}}$, $a_2 \bullet \mathrm{EMG}_{2\mathrm{EMG}} = y_{2\mathrm{EMG}}$, $a_{3\mathrm{EMG}} \bullet \mathrm{EMG}_3 = y_{3\mathrm{EMG}}$, $a_{4\mathrm{EMG}} \bullet \mathrm{EMG}_4 = y_{4\mathrm{EMG}}$.

The results of linear multi-regression (6) had been calculated for four, three, and two cases of multi-regressions. Considering the results of these multi-regressions, we took into account only statistically significant results with coefficients of determination (R^2) that were no less than 0.75 ($R^2 \geq 0.75$). Next, we assessed separate percentage participations of each chosen muscle activation y_{jEMG} with respect to the X, Y, and Z coordinate in each tested window for ***EMG-kinematic relations*** ($part_{y_{iEMG_{WJx}}}$, $part_{y_{iEMG_{WJy}}}$, $part_{y_{iEMG_{WJz}}}$) and ***joint-dynamic relations*** ($part_{y_{iEMG\_COMx}}$, $part_{y_{iEMG\_COMy}}$, $part_{y_{iEMG\_COMz}}$) by using following formulas:

$$part_{y_{iEMG_{WJdim}}} = \frac{y_{iEMG_{WJdim}}}{\sum y_{iEMG_{WJdim}}} \bullet 100\%, \tag{7}$$

$$part_{y_{iEMG\_COM_{dim}}} = \frac{y_{iEMG\_COM_{dim}}}{\sum y_{iEMG\_COM_{dim}}} \bullet 100\%, \tag{8}$$

where dim denotes the *x*-th, *y*-th, and *z*-th components.

Based on results obtained from (7) and (8) formulas we calculated

the value of ***EMG-kinematic relations of accumulated EMG synergy for each muscle activation over time*** y_{jEMG}:

$$accumulated_{y_{iEMG\_kinematic}} = \frac{1}{3} \bullet \left(\left| part_{y_{iEMG\_WJx}} \right| + \left| part_{y_{iEMG\_WJy}} \right| + \left| part_{y_{iEMG\_WJz}} \right| \right), \quad (9)$$

and the value of EMG-dynamic relations of accumulated EMG synergy for each muscle activation over time y_{jEMG}:

$$accumulated_{y_{iEMG\_dynamic}} = \frac{1}{3} \bullet \left(\left| part_{y_{iEMG\_COMx}} \right| + \left| part_{y_{iEMG\_COMy}} \right| + \left| part_{y_{iEMG\_COMz}} \right| \right). \quad (10)$$

Participations obtained from formulas (Eq. (2) along with Eq. (3)) or (Eq. (7) along with Eq. (8)) can be positive (synergistic), negative (antagonistic), or zero. These different signs manifest different directions of a tested action. In this study, we decided to not consider the signs of actions and simplify multidimensional analysis to the average results with respect to the X, Y, and Z coordinates (Eq. (4) - Eq. (5) and Eq. (9)- Eq. (10).

Statistical analyses along with linear piecewise multi-regression analysis were performed in STATISTICA and MATLAB (statistics toolbox) by implementing own scripts. Assuming a threshold of statistical significance ($p = 0.05$), ***the accumulated joint-kinematic synergy of four joint angles over time*** and ***accumulated EMG synergy of four superficial muscles over time*** was assessed by using Bonferroni correction and only considering statistically significant results of multi-regression Eq. (1) and Eq. (6) that simultaneously satisfied two main criteria: 1) coefficients of determination (R^2) were no less than 0.75 ($R^2 \geq 0.75$); 2) analysis of residuals manifested that errors of multi-regression were independent, normally distributed and had equal variances.

3. Results

Results of upper limb motor function of patients with DMD are presented by considering Brooke scale scores: 1) DMD1 (Brooke 3); 2) DMD2 (Brooke 4); 3) DMD3, DMD4, DMD5 (Brooke 5). Also, an analysis of results was conducted by comparing the results of patients with the results of the Control.

3.1. Kinematic synergy of two joint angles over time

Considering all tested trials performed by the Control and each patient with DMD, we presented CC results higher **than 0.4 (this value was treated as a threshold of similarity)** in Fig. 2 and Fig. S2 (Supplement, section S.2). In the supplement we also put Tables S1 – S8 described detailed CC results calculated for all tested subjects and all tested motions. **The impact of CC results (kinematic synergy between each pair of tested joints) was assessed by using the following scoring: the highest (more than 0.9), high (0.7; 0.9], medium (0.5; 0.7], and low [0.4; 0.5]**. Analyzing results related to the vertical motions V1 and V2 (Fig. 2a,b) and complex motion V3 (Fig. 2c), we found following kinematic synergy between each pair of tested joints in all three motions: 1) Control (0.91 ≤ CC) and DMD2 (0.96 ≤ CC) had the highest similarity between SFE-SAA; 2) DMD3 had high similarity between SRot-SAA (0.82 ≤ CC). However, DMD5 had only one high similarity between EFE-SRot (0.77 ≤ CC) in motions V2 and V3.

Considering CC results of horizontal motions H1 and H2 (lightweight) (Fig. S2), we identified that: 1) the highest similarity between SRot-SAA had Control (0.99 ≤ CC), DMD2 (0.94 ≤ CC), DMD3 (0.96 ≤ CC), DMD4 (0.93 ≤ CC), and DMD5 (0.98 ≤ CC); 2) a high similarity between EFE-SRot and EFE-SAA had DMD2 and DMD5; 3) a low-to-high similarity between SRot-SFE, SRot-SAA, SFE-SAA had DMD3.

CC results related to horizontal motions H3 and H4 (heavyweight) (Fig. S2) revealed: 1) similarity between SRot-SAA in Control (0.99 ≤ CC), DMD1 (0.88 ≤ CC), DMD2 (0.98 ≤ CC), DMD4 (0.53 ≤ CC), and DMD5 (0.89 ≤ CC). Also, we found that DMD5 had a similarity between EFE-SRot (0.75 ≤ CC) and DMD4 between EFE-SAA (0.56 ≤ CC). Moreover, CC results of horizontal motion H5 (Fig. S2) point out that in all trials kinematic synergies were between SRot-SAA in Control, DMD1, DMD2, DMD3, and DMD5. Also, in this motion H5 both DMD2 and DMD5 had two similarities between EFE-SRot and EFE-SAA.

3.2. Accumulated joint synergy of four joint angles over time

An accumulated kinematic synergy of four joint angles over time describes contributions of tested joint angles in: 1) joint-kinematic relations (tested joint angles are correlated with the displacement of the wrist joint, i.e., WJx, WJy, WJz); 2) joint-dynamic relations (tested joint angles are correlated with COM of the tested upper limb, i.e., COMx, COMy, COMz). These accumulated results are calculated from distributed results by using formulas (Eq. (4)) and (Eq. (5)). To visualize these distributed results, in Fig. 3A-3B there are given two chosen results for the Control and DMD1 estimated in motion V1. In this study results of accumulated kinematic synergy are presented as accumulated four parts ($a_1 \bullet$ EFE $= y_1 =$ E1, $a_2 \bullet$ SRot $= y_2 =$ S1, $a_3 \bullet$ SFE $= y_3 =$ S2, $a_4 \bullet$ SAA $= y_4 =$ S3) without considering the sign of actions (Fig. 4A-4B).

Results of the accumulated joint synergy of four joint angles over time (Fig. 4A –4B) revealed that:

1. Compared to the Control, the contributions of E1 (related to EFE) in advanced-stage DMD are significantly higher in performing both horizontal and vertical movements. Additionally, a significant predominance of kinematic control over dynamic control was observed in both horizontal and vertical movements.
2. Compared to the Control, the contributions of S1 (related to SRot) in advanced-stage DMD decrease significantly in horizontal movements and increase in vertical movements. Additionally, a slight

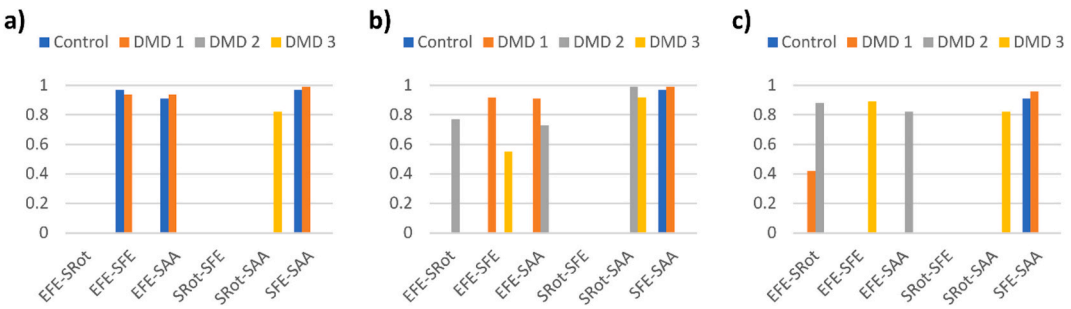

Fig. 2. Results of CC coefficients (vertical axis describes the CC coefficient): (a) Motion V1, (b) Motion V2, (c) Motion V3.

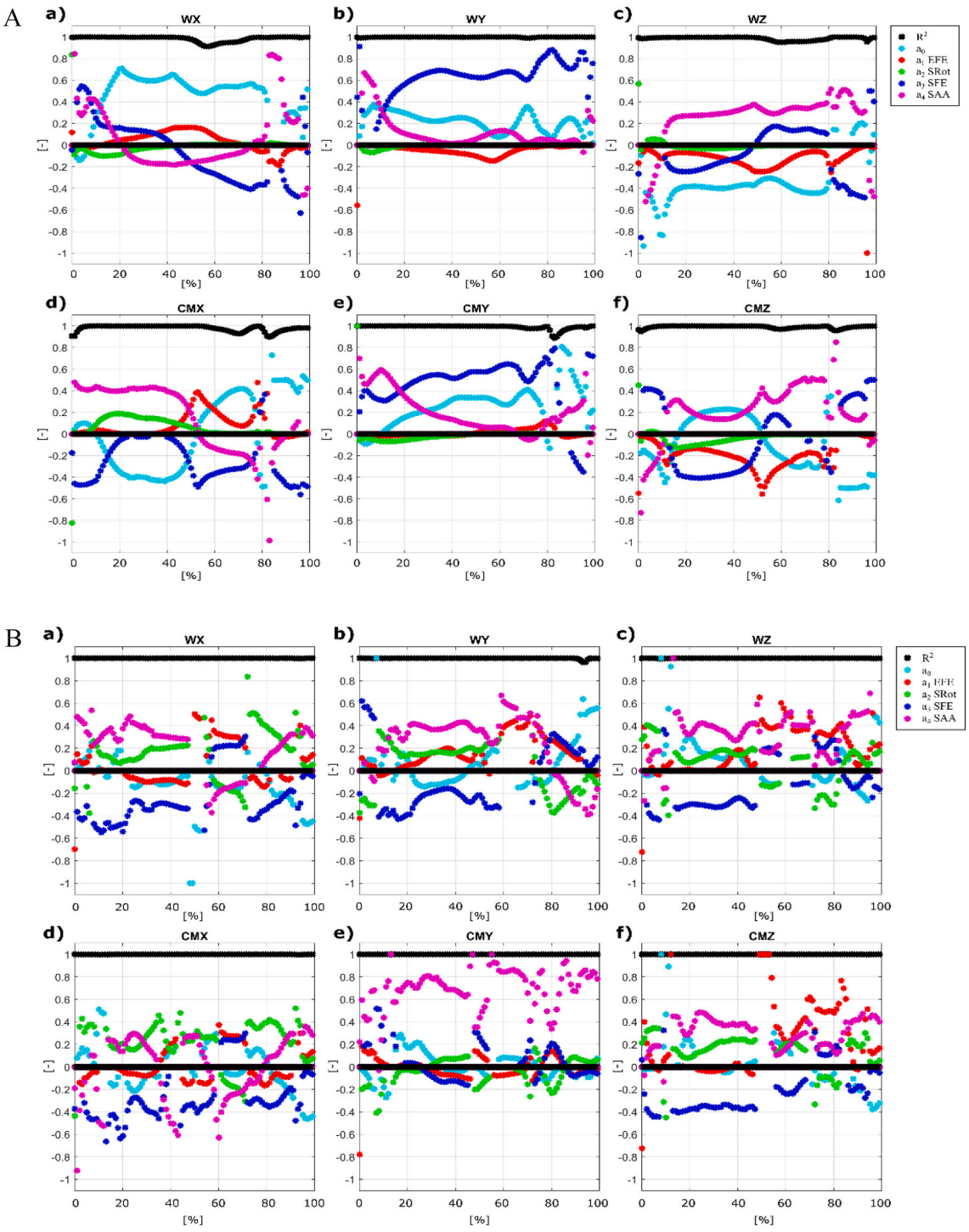

Fig. 3. A. Results of contributions of tested joint angles of Control over time in motion V1 for kinematic relations: (**a**) WX, (**b**) WY, (**c**) WZ; dynamic relations: (**d**) CMX, (**e**) CMY, (**f**) CMZ.
B. Results of contributions of tested joint angles of DMD1 over time in motion V1 for kinematic relations: (**a**) WX, (**b**) WY, (**c**) WZ; dynamic relations: (**d**) CMX, (**e**) CMY, (**f**) CMZ.

predominance of dynamic control over kinematic control was observed in both horizontal and vertical movements.

3. Compared to the Control, the contributions of S2 (related to SFE) in advanced-stage DMD decrease significantly in vertical movements. Additionally, a slight predominance of dynamic control over kinematic control was observed in both horizontal and vertical movements.
4. Compared to the Control, the contributions of S3 (related to SAA) in advanced-stage DMD (DMD4 and DMD5) decrease in horizontal movements, except DMD3. Additionally, a slight predominance of dynamic control over kinematic control was observed in both horizontal and vertical movements.

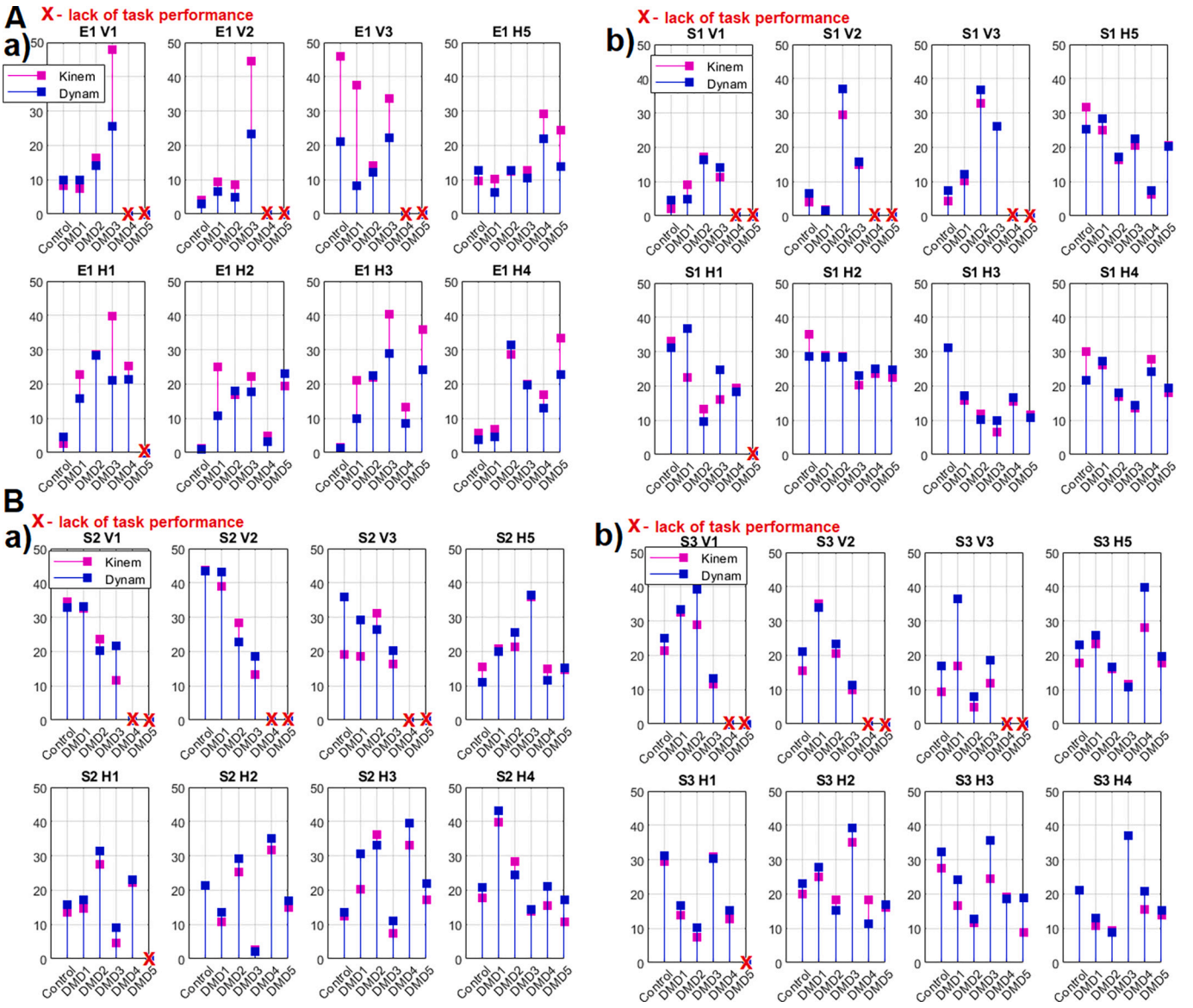

Fig. 4. A. Results of the accumulated joint synergy of four joint angles over time (joint-kinematic (Kinem) and joint-dynamic relations (Dynam)) for the joint: (**a**) E1 (EFE), (**b**) S1 (SRot).
B. Results of the accumulated joint synergy of four joint angles over time (joint-kinematic (Kinem) and joint-dynamic relations (Dynam)) for the joint: (**a**) S2 (SFE), (**b**) S3 (SAA).

3.3. Predominant type of control of accumulated joint synergy of four joint angles over time

Results of predominant types of control of accumulated kinematic synergy of four joint angles over time are presented in Fig. 5 for motion: V1 (Fig. 5a), V2 (Fig. 5b), V3 (Fig. 5c), H1 (Fig. 5d), H2 (Fig. 5e), H3 (Fig. 5f), H4 (Fig. 5g), and H5 (Fig. 5h). Note that the Y-direction is the vertical axis that is related to the gravity influence, thus X and Z directions refer to the horizontal plane. In all tested motions each tested patient with DMD controlled all joint-kinematic relations (WX, WY, WZ) and all joint-dynamic relations (CMX, CMY, CMZ) in all directions (X, Y, Z). These results are very different in comparison with the healthy Control, which controlled with a very high predominant types of control ($R^2 \geq 0.98$): a) all joint-kinematic and joint-dynamic relations in vertical motion V1 (Fig. 10a) and horizontal motion H4 (Fig. 5g); b) all joint-dynamic relations and two joint-kinematic relations (WY and WZ) in vertical motion V2 (Fig. 5b); c) one joint-dynamic relation (CMY) and two joint-kinematic relations (WY and WZ) in motion V3 (Fig. 5c); d) two joint-dynamic relations (CMX, CMZ) and one joint-kinematic relation (WX) in motion H1 (Fig. 5d); e) all joint-dynamic relations and two joint-kinematic relations (WX, WY) in motion H2 (Fig. 5e); f) one joint-kinematic relation (WY) in motion H3 (Fig. 5f); g) one joint-dynamic relation (CMY) in motion H5 (Fig. 5h).

3.4. Accumulated EMG synergy of four superficial muscles over time

To assess an accumulated EMG synergy of four superficial muscles over time (EMG1, EMG2, EMG3, and EMG4) and identify ***EMG-kinematic relations*** (activity of tested muscles are correlated with the displacement of the wrist joint) along with ***EMG-dynamic relations*** (activity of tested muscles are correlated with COM), first we identified which activations of tested muscle in this study influenced examined motions. To perform this identification, we calculated the mean value of each processed EMG (RMS EMG) for each tested track by considering the EMG threshold (onset) (described in Section 2). Next, two clinicians performed visual inspections of processed EMGs and motion tracks and

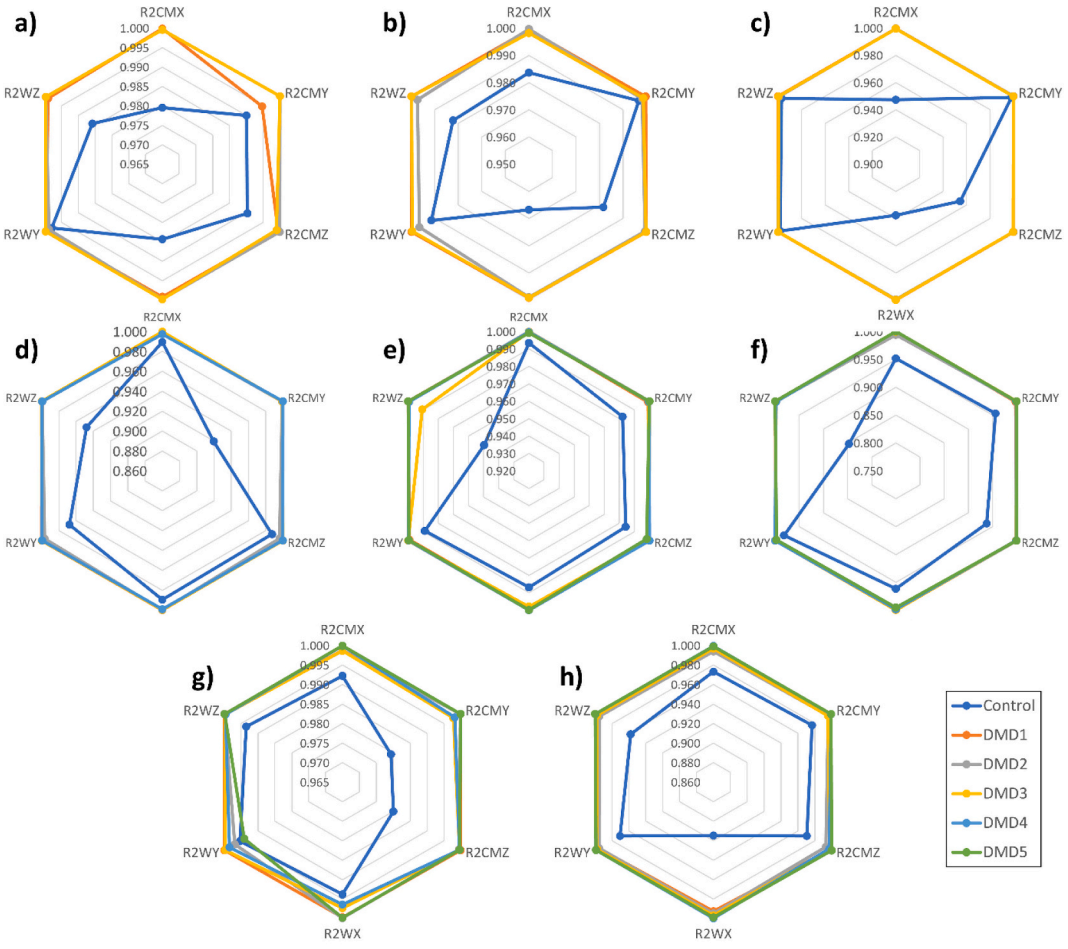

Fig. 5. Results of predominant types of control of accumulated kinematic synergy of four joint angles over time for motion: (**a**) V1, (**b**) V2, (**c**) V3, (**d**) H1, (**e**) H2, (**f**) H3, (**g**) H4, (**h**) H5.

assumed that 0.04 of the mean value of each processed EMG (RMS EMG) should be treated as a threshold to consider EMG data in the linear multi-regression procedures (Eq. (6)) to assess ***EMG-kinematic relations*** and ***EMG-dynamics relations*** according to formulas Eq. (9) and Eq. (10).

In Fig. 6 along with Supplement section S.3, there are given mean values of each processed EMG (EMG1, EMG2, EMG3, EMG4). Analysis of the results reveals that:

1. performing horizontal movements (H1, H2, H3, H4), a healthy individual (Control) uses only EMG3, whereas weaker patients with DMD activate all examined muscles (EMG1, EMG2, EMG3, EMG4);
2. performing vertical movements (V1, V2, V3), a healthy individual (Control) and DMD1 use all tested muscles (EMG1, EMG2, EMG3, EMG4), whereas DMD2 and DMD3 or use only selected muscles.

Results of accumulated EMG synergy of four superficial muscles over time (EMG1, EMG2, EMG3, and EMG4) are given in Fig. 7A–7B. These results revealed that:

1. Contributions of trapezius, triceps, biceps, and deltoid in vertical and horizontal motions are higher in patients with DMD than in Control. The predominance of dynamic control is visible in horizontal and vertical motions.
2. Compared to the Control, the contributions of EMG1 (Trapezius) in weaker patients with DMD are significantly higher in horizontal movements and slightly higher in vertical movements.
3. Compared to the Control, the contributions of EMG4 (Biceps) in patients with DMD are higher in both horizontal and vertical movements with a predominance of kinematic control in horizontal movements with lighter weights and a predominance of dynamic control with heavier weights.

4. Discussion

This study presents a novel scoring, which is composed of four indexes: three kinematic indexes and one related to EMG data. In this multi-case study, the results are assessed for five patients with DMD at different stages (Brooke 3–5) of the disease progression, age (14.3 ± 1.8 years), and one healthy adolescent (Control), which anthropometric proportions were similar to the tested patients. All tests of functional tasks were performed in clinical conditions by using a motion capture system and EMG system that had synchronously collected data during testing. Presented novel scoring implies: 1) kinematic synergy of two joint angles over time; 2) accumulated joint synergy of four joint angles over time (relation A); 3) predominant type of control of accumulated joint synergy of four joint angles over time; 4) accumulated EMG synergy of four superficial muscles over time (relation B). An analysis of all results was conducted by considering the disease progression of the patients scored on the base of the Brooke scale.

Due to the fact that all tests conducted in this study had been performed without any preliminary training, there are some differences in the motion performance of each patient. These differences point out that patients with DMD tried to use different motor patterns to perform the same motion.

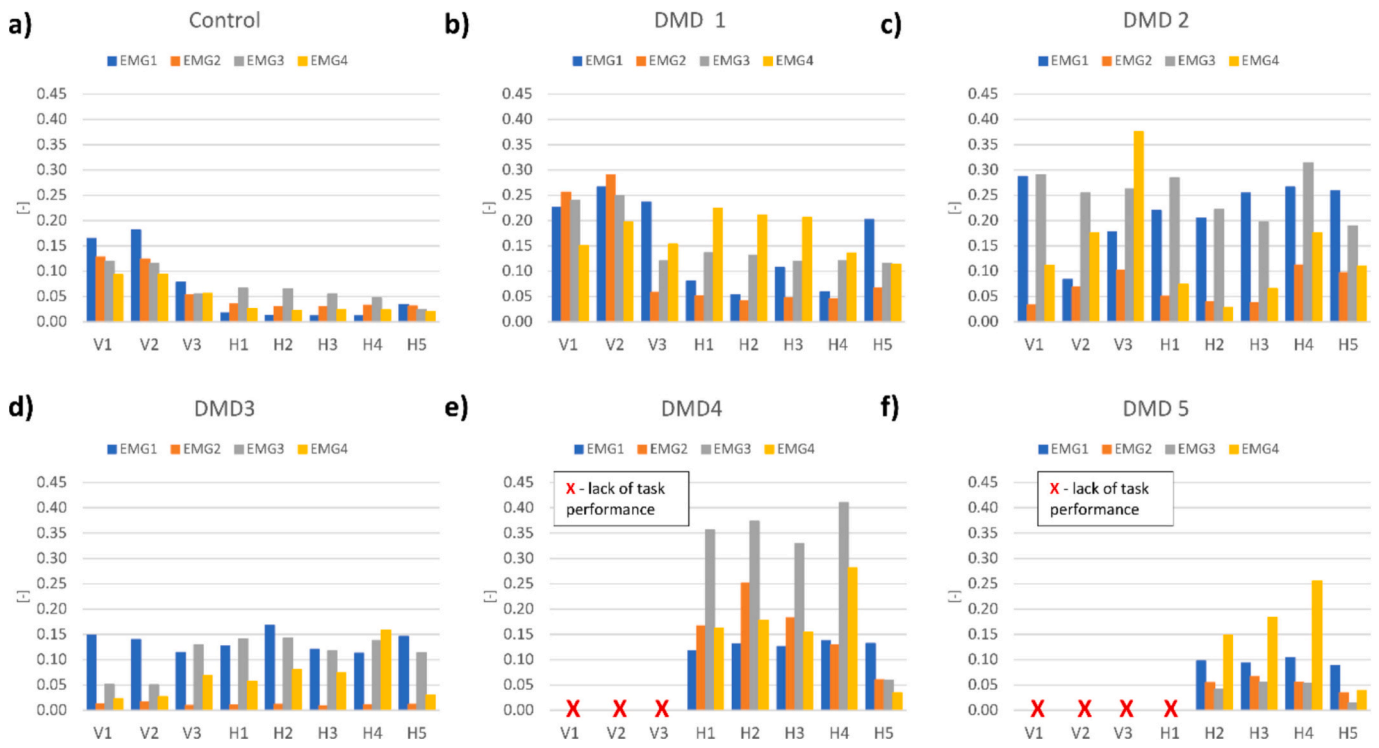

Fig. 6. Mean values of each processed EMG (RMS EMG).

4.1. *Kinematic synergy of two joint angles over time*

Analyzing the results of ***the* kinematic synergy of two joint angles over time** (CC results (Section 3.1)), it is possible to identify two joints, in which two chosen joint angles are changing at the same time. Moreover, assessment of this synergy can reveal kinematic changes in motion performance. In this study we found that in tested vertical motions V1 and V2 along with a complex motion V3 (Fig. 2a, 2b, 2c): 1) Control and DMD2 had three the highest synergies (SFE-SAA, EFE-SAA, EFE-SFE) in motion V1 and only one synergy related to SFE-SAA in motions V2 and V3; 2) DMD2 and DMD5 had only the highest synergy related to EFE-SAA in motion V2 and low-high synergy related to EFE-SRot in motion V3; 3) DMD3 and DMD5 had only one similar synergy related to SRot-SAA in motion V2. Concerning tested horizontal motions H1 and H2 (lightweight) we found that (Fig. S2a, S2b): 1) Control and four tested patients with DMD (except DMD1) had the highest synergy related to SRot-SAA; 2) Control, DMD2, DMD4, and DMD5 had synergy related to EFE-SRot and EFE-SAA in motion H1; 3) four patients with DMD (except DMD4) had synergy related to EFE-SRot while DMD2 and DMD5 had synergy related to EFE-SAA in motion H2; 4) DMD3 had synergy related SRot-SFE and SFE-SAA in motions H1 and H2.

In tested horizontal motions with the heavyweight we revealed that (Fig. S2c, S2d): 1) four patients with DMD (except DMD3) and Control had synergy related to SRot-SAA in motion H3, however, all tested patients with DMD and Control had synergy related to SRot-SAA in motion H4; 2) three patients (DMD1, DMD2, and DMD5) had synergy related to EFE-SRot and EFE-SAA in motion H3; 3) there were observed different behaviors in motion H4, i.e., DMD3 had synergy related to SRot-SFE and SFE-SAA, DMD4 had synergy related to EFE-SAA, while DMD5 had synergy related to EFE-SRot. On the other hand, in a tested horizontal complex motion (following the track by a finger) (Fig. S2e): 1) a Control and four tested patients with DMD (except DMD4) had synergy related to SRot-SAA; 2) DMD2 and DMD5 had synergy related to EFE-SRot and EFE-SAA, while DMD4 had synergy related to SFE-SAA.

It is important to note that visual inspection of the recorded motions of examined subjects does not always allow for the identification of compensatory motions. Therefore, in clinical practice, CC results can help reveal kinematic synergies and determine whether the tested patient is using (or he/she has just started using) a compensation strategy to perform a motion.

4.2. *Accumulated joint synergy of four joint angles over time (relation A)*

Results presented in Section 3.2 can be used to detect contributions of tested joint angles (EJ, SRot, SEF, SAA) in the global task performance, which is correlated with the displacement of the tested wrist joint and/or the displacement of the COM of the tested upper limb.

Results presented in Fig. 4A – 4B reveal that: a) in all vertical motions (V1, V2, V3) weaker patients with DMD performed more EFE motion by using more kinematic-joint relations; b) with respect to the healthy Control, these weaker patients with DMD had decreased SFE motion and performed more SRot motion along with SAA motion (that might be caused by a compensation motion of a trunk). However, DMD3 performed SAA motion in a minimum way in comparison with DMD patients with better function. Moreover, this DMD3 was the only patient with a Brooke 5 score that still could lift the upper limb above the table surface (against gravity force).

In the results of horizontal motions (H1, H2, H3, H4) (Fig. 4A – 4B) we can see that: 1) all patients with DMD performed extensive EFE motion in comparison to the healthy Control (that performed very small EFE motion in all horizontal tests); 2) for lighter weights (H1,H2,H3) almost all patients with DMD performed EFE motion with more kinematic-joint relations, moreover, these relations were identified in the weakest patients with DMD in tests with the heaviest weight (H4); 3) weaker patients with DMD performed less SRot motion; 4) almost all patients with DMD along with the healthy Control performed SFE motion by mainly using dynamic-joint relations with respect to kinematic-joint relations (except DMD2 that used kinematic-joint relations in tests with the heaviest weights); 5) all tested subjects used SAA motion in all horizontal tests with nearly the same amount of kinematic-joint relations and dynamic-joint relations. Moreover, all these subjects performed SAA motion in tests with the heaviest weights mainly by using

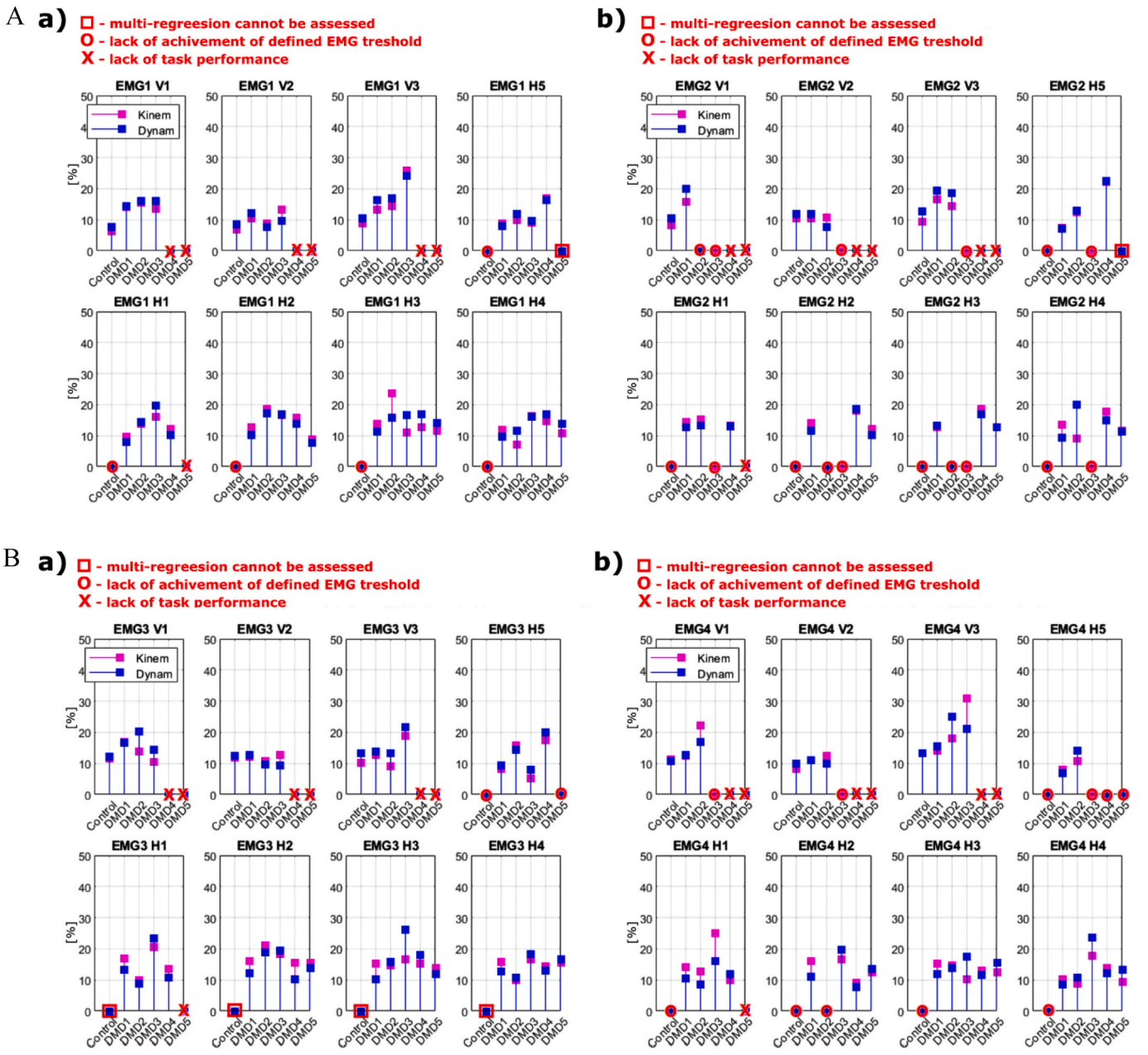

Fig. 7. A. Results of accumulated EMG synergy of four superficial over time (joint-kinematic (Kinem) and joint-dynamic relations (Dynam)) related to a) EMG1 and b) EMG2.
B. Results of accumulated EMG synergy of four superficial over time (joint-kinematic (Kinem) and joint-dynamic relations (Dynam)) related to a) EMG3 and b) EMG4.

dynamic-joint relations, however, DMD2 used kinematic-joint relations in tests with the heaviest weights.

In the complex horizontal motion (H5) (Fig. 4A – 4B) we found that in comparison to the healthy Control weaker patients with DMD: 1) performed more EFE motion by using kinematic-joint relations; 2) performed less SRot motion by using nearly the same amount of kinematic-joint and dynamic-joint relations.

Our findings revealed a greater contribution of EFE motion in the performance of tested patients with DMD, especially those in weaker health conditions. This can be explained by the fact that in the whole kinematic chain, the complex motions are performed by using motor synergies. As patients with DMD first lose these synergies in proximal muscles (caused by a progression of muscle weakening) the contribution of EFE motion becomes greater.

Regarding the issue related to the active functional ROM of each tested subject, we have included the results in the Supplement (section S.1). Comparing these results with published ones (Janssen et al., 2017), it is important to stress that in this study we assessed the results of active functional ROM in functional ADL tasks. We found that in vertical motions (V1 and V2) (Figs. S1Aa, S1Ab), the range of SFE and the range of SAA show a decreasing trend in the results of DMD1 (Brooke3), DMD2 (Brooke4), and DMD3 (Brooke5). This trend is consistent with the results of 'Shoulder flexion' given in (Janssen et al., 2017). However, the range of EFE in complex vertical motion V3 (Fig. S1B) and the range of SRot in horizontal motions H1, H2, H3, and H4 (Fig. S1C) do not show the same decreasing trend for the tested patients with DMD as reported for 'Elbow flexion' and 'Shoulder adduction (in the horizontal plane)' in the study (Janssen et al., 2017). This discrepancy can be explained by the fact that the active ROM results presented in (Janssen et al., 2017) and the active functional ROM presented in this study are performed with different configurations of the upper limb segments with respect to the trunk. Moreover, the high level of standard deviation observed in

some tested functional motions aligns with evidence given in papers (Bertomeu-Motos et al., 2017; Magermans et al., 2005), which point out that a high level of standard deviation in trajectories and joint angles is observed in the performance of healthy subjects in each simple ADL motion.

4.3. Predominant type of control of accumulated joint synergy of four joint angles over time

The results of predominant types of control of accumulated kinematic synergy of four joint angles over time (Section 3.3) revealed the differences in types of control between Control subject and tested patients with DMD. Presented results are in line with observations described in (Brooke et al., 1989) that revealed an increase in upper limb motor compensatory strategies, which can vary at each stage of the disease progression and with different activities, and each patient with DMD may present a unique evolution of activity performance within a motor pattern.

4.4. Accumulated EMG synergy of four superficial muscles over time (relation B)

Results presented in Section 3.4 allow the clinicians to estimate the contributions of tested superficial muscles (Trapezius (EMG1), Lateral Triceps Brachii (EMG2), Deltoideus Anterior (EMG3), and Biceps Brachii (EMG4)) in the global task performance, which is correlated with the displacement of the tested wrist joint and/or the displacement of the COM of the tested upper limb. These results of accumulated EMG synergy were assessed from statistically significant EMG-kinematic relations and EMG-dynamic relations.

Considering processed EMGs (RMS EMGs), that had been used to assess accumulated EMG synergy, it is worth noting that the following percentage of four tested muscles exceeded the assumed threshold for RMS EMGs in tested motions: healthy Control – 50 %, DMD1–100 %, DMD2–87.5 %, DMD3–65.6 %, DMD4–95.0 % (in 62.5 % of all motions tested in this study), DMD5–81.2 % (in 50.0 % of all motions tested in this study). These results along with results of accumulated EMG synergy (Section 3.4) reveal that patients with DMD used muscles in an excessive way with respect to the Control in tested functional activities. This excessive way indicates that tested motions were difficult to perform for these patients. Moreover, this excessive muscle activation causes the patient's fatigability in daily activities. These findings are similar to the observations described by Janssen (Janssen et al., 2017), who revealed increased normalized EMG amplitudes of upper limb muscles in patients with DMD even in the early stage of the disease in comparison to healthy controls.

The results of accumulated EMG synergy showed: 1) Lateral Triceps Brachii (EMG2) did not take part in the movement of the weakest patient with DMD that was able to perform vertical motion (DMD3), although this muscle was used to perform most of the tested horizontal motions by DMD4 and DMD5; 2) Trapezius (EMG1) contributed in shoulder lifting of tested patients with DMD and this lifting should be treated as a manifestation of the most common compensation observed in reaching and lifting tasks. Described observations are similar to those presented by Essers who assessed functional tasks in patients with facioscapulohumeral dystrophy (Essers et al., 2023) and revealed that muscle coordination of the upper limb of these patients was altered and less consistent compared with healthy controls.

4.5. Limitations

The study is not free from limitations. Firstly, we tested a small number of volunteers because DMD is a rare disease, and we collected a group of wheelchair patients at a narrow age range (15.2 ± 2.2 years). Secondly, the study was conducted in clinical conditions, as it was not possible to test patients with DMD in laboratory conditions and to align them in a laboratory chair due to trunk weakness. Thirdly, muscle weakness and contracture influenced the trajectory of the movements of tested patients. That is why tested motions, that were spontaneous natural motions without any corrections concerning the trajectory and pace, could not be repeated in the same way. Fourthly, the Noraxon EMG measurement system used in this study only could collect the data within the [10,500] Hz frequency range. This technical limitation did not allow us to consider EMG data within the larger spectrum of frequency. Fifthly, the seated position (in a wheelchair) of weaker patients (being in more advanced stages of the disease) was more asymmetrical and led to emerge compensatory movements through trunk rotation. Moreover, trunk movements were not taken into consideration in the scope of this study. That is why further studies should be designed to consider the influence of trunk compensatory movement and trunk muscle activity while performing tested activities by using the upper limb. For a better insight into the model of the upper limb movement patterns in DMD progression, further studies on larger groups in different functional stages are required with the comparison to a representative healthy control group.

5. Conclusions

The aim of this study was to propose a novel scoring to analyze kinematic patterns and EMG patterns of ADL functional motions of patients with DMD. The proposed scoring involves four indexes (three kinematic indexes and one EMG index) that can help clinicians assess the whole tracks of tested motions performed by the tested patients. In the scope of this study, we tested patients with DMD who used wheelchairs and were at different stages of DMD progression. Tested patients performed ADL functional tasks in clinical conditions. Presented novel scoring can help identify the movement coordination and gradually appearing compensations of the upper limb of the patient with DMD caused by a progression of the disease. Moreover, the proposed scoring allows obtaining information about changes in inter-joint coordination and changes in compensatory movement. It is worth noting that the proposed novel scoring used non-invasive testing that is easy to get on with patients with DMD, especially for weaker patients with muscle atrophy, joint contractures, and diminished ROM. Moreover, this proposed protocol of testing could be easily implemented in clinical conditions to perform ADL testing.

To our knowledge, the findings presented have not been reported to date. That is why these results can fill the gap related to the mobility of patients with DMD being in different stages of disease progression. Moreover, the kinematic synergy of two joint angles and the accumulated kinematic synergy of four joint angles along with the accumulated EMG synergy of four tested superficial muscles reflect the coordination of tested joints, which is directly related to the activity of muscles at the tested joints. These accumulated results allow assessing the participation of tested joints and tested muscles in each tested ADL activity in a given condition by specifying the types of relation (kinematic control or dynamic control) that could be associated with the senses in the following way: 1) kinematic control can be performed by using information from proprioception, visual and tactile senses; 2) dynamic control is mainly performed on the base of information from proprioception and tactile senses, i.e. without information from a visual sense. Moreover, the findings described in this paper can be compared with the performance of the healthy subject, who coordinates his/her motion through joints and muscles cooperation in the normal conditions in the following way (the sign is put along with a joint to specify the direction of the motion): 1) elbow flexion (+E1) - Biceps Brachii; 2) elbow extension (−E1) -Triceps Brachii; 3) shoulder internal rotation (+S1) - Subscapularis, Teres Major, Latissimus Dorsi, and Pectoralis Major; 4) shoulder external rotation (—S1) - Teres Minor and Infraspinatus; 5) shoulder flexion (+S2) - Anterior Portion of Deltoid; 6) shoulder extension (—S2) - Triceps Brachii, Posterior Portion of Teres Major Deltoid and Latissimus Dorsi; 7) shoulder abduction (+S3) – Deltoid,

Supraspinatus; 8) shoulder adduction (—S3) - Pectorals Major.

Presented findings should be treated as the first insight into the movement patterns of the upper limb in patients with DMD, and these findings should be interpreted with caution due to the small sample size.

Results presented in this multi-case study revealed differences in movement patterns in comparison to the chosen healthy Control that has anthropometric proportions similar to the tested patients with DMD. Furthermore, the described data does not define the unequivocal course of muscle wasting. Reported diversity in kinematic data among patients is caused by the redundancy of the muscular system and individual compensatory habits. To reveal changes in kinematic patterns and superficial EMG patterns along with the development of compensations in muscular dystrophies over time a long-term follow-up is needed to be conducted on bigger groups of patients with DMD.

CRediT authorship contribution statement

Agnieszka Sobierajska-Rek: Conceptualization, Funding acquisition, Investigation, Methodology, Project administration, Resources, Validation, Writing – original draft, Writing – review & editing. **Wiktoria Wojnicz:** Conceptualization, Data curation, Formal analysis, Methodology, Software, Writing – original draft, Writing – review & editing. **Bartłomiej Zagrodny:** Data curation, Supervision, Writing – original draft, Writing – review & editing. **Michał Ludwicki:** Data curation, Visualization, Writing – original draft, Writing – review & editing. **Katarzyna Pytka:** Investigation. **Joanna Jabłońska-Brudło:** Investigation, Resources.

Informed consent statement

Written informed consent was obtained from all subjects involved in the study.

Institutional review board statement

The study was conducted in accordance with the Declaration of Helsinki and approved by the Institutional Review Board (or Ethics Committee) of the Ethic Committee of Medical University of Gdansk NKBBN/23/2019, NKBBN/23–708/2019, NKBBN/23–409/2020.

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Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.clinbiomech.2025.106542.

Data availability

The datasets used in the study are available from the corresponding author upon reasonable request.

References

Alison, B., Elizabeth, G., Steve, L., Andrew, B., Guy, M., Volker, S., 2012. Animal models of Duchenne muscular dystrophy, with special reference to the mdx mouse. Biocyber. Biomed. Eng. 32, 3–15. https://doi.org/10.1016/S0208-5216(12)70045-1.

Alt Murphy, M., Willén, C., Sunnerhagen, K.S., 2011. Kinematic variables quantifying upper-extremity performance after stroke during reaching and drinking from a glass. Neurorehabil. Neural Repair 25, 71–80. https://doi.org/10.1177/1545968310370748.

Attias, M., Chevalley, O., Bonnefoy-Mazure, A., De Coulon, G., Cheze, L., Armand, S., 2016. Effects of contracture on gait kinematics: a systematic review. Clin. Biomech. 33, 103–110. https://doi.org/10.1016/j.clinbiomech.2016.02.017.

Barański, R., Wojnicz, W., Zagrodny, B., Ludwicki, M., Sobierajska-Rek, A., 2024. Towards hand grip force assessment by using EMG estimators. Measurement 226, 114137. https://doi.org/10.1016/j.measurement.2024.114137.

Bendixen, R.M., Morgenroth, L.P., Clinard, K.L., 2016. Engaging participants in rare disease research: a qualitative study of Duchenne muscular dystrophy. Clin. Ther. 38, 1474–1484.e2. https://doi.org/10.1016/j.clinthera.2016.04.001.

Bertomeu-Motos, A., Delegido, I., Ezquerro, S., Lledó, L.D., Catalan, J.M., Garcia-Aracil, N., 2017. Upper-limb motion analysis in daily activities using wireless inertial sensors. In: Ibáñez, J., González-Vargas, J., Azorín, J.M., Akay, M., Pons, J.L. (Eds.), Converging Clinical and Engineering Research on Neurorehabilitation II, vol. 15. Springer International Publishing, Cham, pp. 1079–1083. https://doi.org/10.1007/978-3-319-46669-9_175.

Birnkrant, D.J., Bushby, K., Bann, C.M., Alman, B.A., Apkon, S.D., Blackwell, A., et al., 2018. Diagnosis and management of Duchenne muscular dystrophy, part 2: respiratory, cardiac, bone health, and orthopaedic management. Lancet Neurol. 17, 347–361. https://doi.org/10.1016/S1474-4422(18)30025-5.

Brooke, M.H., Fenichel, G.M., Griggs, R.C., Mendell, J.R., Moxley, R., Florence, J., et al., 1989. Duchenne muscular dystrophy: patterns of clinical progression and effects of supportive therapy. Neurology 39, 475. https://doi.org/10.1212/WNL.39.4.475.

Carpinella, I., Lencioni, T., Bowman, T., Bertoni, R., Turolla, A., Ferrarin, M., et al., 2020. Effects of robot therapy on upper body kinematics and arm function in persons post stroke: a pilot randomized controlled trial. J. NeuroEng. Rehabil. 17, 10. https://doi.org/10.1186/s12984-020-0646-1.

Case, L.E., Apkon, S.D., Eagle, M., Gulyas, A., Juel, L., Matthews, D., et al., 2018. Rehabilitation management of the patient with Duchenne muscular dystrophy. Pediatrics 142, S17–S33. https://doi.org/10.1542/peds.2018-0333D.

Centen, A., Lowrey, C.R., Scott, S.H., Yeh, T.-T., Mochizuki, G., 2017. KAPS (kinematic assessment of passive stretch): a tool to assess elbow flexor and extensor spasticity after stroke using a robotic exoskeleton. J. NeuroEng. Rehabil. 14, 59. https://doi.org/10.1186/s12984-017-0272-8.

Connolly, A.M., Malkus, E.C., Mendell, J.R., Flanigan, K.M., Miller, J.P., Schierbecker, J. R., et al., 2015. Outcome reliability in non-ambulatory boys/men with duchenne muscular dystrophy: corticosteroids in boys/men with DMD. Muscle Nerve 51, 522–532. https://doi.org/10.1002/mus.24346.

De Souza, M.A., Figueiredo, M.M.L., De Baptista, C.R.D.J.A., Aldaves, R.D., Mattiello-Sverzut, A.C., 2016. Beneficial effects of ankle–foot orthosis daytime use on the gait of Duchenne muscular dystrophy patients. Clin. Biomech. 35, 102–110. https://doi.org/10.1016/j.clinbiomech.2016.04.005.

Duan, D., Goemans, N., Takeda, S., Mercuri, E., Aartsma-Rus, A., 2021. Duchenne muscular dystrophy. Nat. Rev. Dis. Primers 7, 13. https://doi.org/10.1038/s41572-021-00248-3.

Eagle, M., Bourke, J., Bullock, R., Gibson, M., Mehta, J., Giddings, D., et al., 2007. Managing Duchenne muscular dystrophy – the additive effect of spinal surgery and home nocturnal ventilation in improving survival. Neuromuscul. Disord. 17, 470–475. https://doi.org/10.1016/j.nmd.2007.03.002.

Ellis, M.D., Schut, I., Dewald, J.P.A., 2017. Flexion synergy overshadows flexor spasticity during reaching in chronic moderate to severe hemiparetic stroke. Clin. Neurophysiol. 128, 1308–1314. https://doi.org/10.1016/j.clinph.2017.04.028.

Essers, J.M.N., Meijer, K., Peters, A.A., Murgia, A., 2023. The effects of facioscapulohumeral dystrophy and dynamic arm support on upper extremity muscle coordination in functional tasks. Neuromuscul. Disord. 33, 651–659. https://doi.org/10.1016/j.nmd.2022.11.002.

Germanotta, M., Cortellini, L., Insalaco, S., Aprile, I., 2023. Effects of upper limb robot-assisted rehabilitation compared with conventional therapy in patients with stroke: preliminary results on a daily task assessed using motion analysis. Sensors 23, 3089. https://doi.org/10.3390/s23063089.

Glickman, N.S., Crump, C., Hamerdinger, S., 2020. Language Deprivation Is a Game Changer for the Clinical Specialty of Deaf Mental Health, p. 54.

Hiebeler, M., Thiele, S., Reilich, P., Bernert, G., Walter, M.C., 2023. Time to diagnosis of Duchenne muscular dystrophy in Austria and Germany. Sci. Rep. 13, 179. https://doi.org/10.1038/s41598-022-27289-2.

Janssen, M.M.H.P., Bergsma, A., Geurts, A.C.H., de Groot, I.J.M., 2014. Patterns of decline in upper limb function of boys and men with DMD: an international survey. J. Neurol. 261, 1269–1288. https://doi.org/10.1007/s00415-014-7316-9.

Janssen, M.M.H.P., Harlaar, J., Koopman, B., de Groot, I.J.M., 2017. Dynamic arm study: quantitative description of upper extremity function and activity of boys and men with duchenne muscular dystrophy. J. NeuroEng. Rehabil. 14, 45. https://doi.org/10.1186/s12984-017-0259-5.

Khallaf, M.E., Ameer, M.A., Fayed, E.E., 2017. Effect of task specific training and wrist-fingers extension splint on hand joints range of motion and function after stroke. NRE 41, 437–444. https://doi.org/10.3233/NRE-162128.

Koeks, Z., Bladen, C.L., Salgado, D., van Zwet, E., Pogoryelova, O., McMacken, G., et al., 2017. Clinical outcomes in Duchenne muscular dystrophy: a study of 5345 patients from the TREAT-NMD DMD global database. JND 4, 293–306. https://doi.org/10.3233/JND-170280.

Magermans, D.J., Chadwick, E.K.J., Veeger, H.E.J., van der Helm, F.C.T., 2005. Requirements for upper extremity motions during activities of daily living. Clin. Biomech. (Bristol, Avon) 20, 591–599. https://doi.org/10.1016/j.clinbiomech.2005.02.006.

Maletsky, L.P., Sun, J., Morton, N.A., 2007. Accuracy of an optical active-marker system to track the relative motion of rigid bodies. J. Biomech. 40, 682–685. https://doi.org/10.1016/j.jbiomech.2006.01.017.

Maria, P., Lydia, K., Jia-Jin, J.C., Irena, H.-P., 2012. Assessment of human motoneuron afterhyperpolarization duration in health and disease. Biocyber. Biomed. Eng. 32, 43–61. https://doi.org/10.1016/S0208-5216(12)70041-4.

Mayhew, A., Mazzone, E.S., Eagle, M., Duong, T., Ash, M., Decostre, V., et al., 2013. Development of the performance of the upper limb module for Duchenne muscular dystrophy. Dev. Med. Child Neurol. 55, 1038–1045. https://doi.org/10.1111/dmcn.12213.

Mayhew, A.G., Coratti, G., Mazzone, E.S., Klingels, K., James, M., Pane, M., et al., 2020. Performance of upper limb module for Duchenne muscular dystrophy. Develop. Med. Child. Neuro. 62, 633–639. https://doi.org/10.1111/dmcn.14361.

Mendell, J.R., Shilling, C., Leslie, N.D., Flanigan, K.M., Al-Dahhak, R., Gastier-Foster, J., et al., 2012. Evidence-based path to newborn screening for duchenne muscular dystrophy. Ann. Neurol. 71, 304–313. https://doi.org/10.1002/ana.23528.

Mercuri, E., Mazzone, E., 2011. Choosing the right clinical outcome measure: from the patient to the statistician and back. Neuromuscul. Disord. 21, 16–19. https://doi.org/10.1016/j.nmd.2010.09.003.

Mercuri, E., McDonald, C., Mayhew, A., Florence, J., Mazzone, E., Bianco, F., et al., 2012. International workshop on assessment of upper limb function in Duchenne muscular dystrophy. Neuromuscul. Disord. 22, 1025–1028. https://doi.org/10.1016/j.nmd.2012.06.006.

Mercuri, E., Bönnemann, C.G., Muntoni, F., 2019. Muscular dystrophies. Lancet 394, 2025–2038. https://doi.org/10.1016/S0140-6736(19)32910-1.

Merletti, R., Cerone, G.L., 2020. Tutorial. Surface EMG detection, conditioning and pre-processing: best practices. J. Electromyogr. Kinesiol. 54, 102440. https://doi.org/10.1016/j.jelekin.2020.102440.

Naarding, K.J., Janssen, M.M.H.P., Boon, R.D., Bank PJM, Matthew, R.P., Kurillo, G., et al., 2022. The black box of technological outcome measures: An example in Duchenne muscular dystrophy. JND 9, 555–569. https://doi.org/10.3233/JND-210767.

Pangalila, R., Bartels, B., Bergen, M., Cobben, N., Stam, H., Roebroeck, M., 2011. Upper limb function in adults with Duchenne muscular dystrophy. J. Rehabil. Med. 43, 770–775. https://doi.org/10.2340/16501977-0841.

Pauk, J., Daunoraviciene, K., Ziziene, J., Minta-Bielecka, K., Dzieciol-Anikiej, Z., 2023. Classification of muscle activity patterns in healthy children using biclustering algorithm. Biomed. Signal Process. Control 84, 104731. https://doi.org/10.1016/j.bspc.2023.104731.

Rashid, U., Niazi, I.K., Signal, N., Farina, D., Taylor, D., 2019. Optimal automatic detection of muscle activation intervals. J. Electromyogr. Kinesiol. 48, 103–111. https://doi.org/10.1016/j.jelekin.2019.06.010.

Ricotti, V., Kadirvelu, B., Selby, V., Festenstein, R., Mercuri, E., Voit, T., et al., 2023. Wearable full-body motion tracking of activities of daily living predicts disease trajectory in Duchenne muscular dystrophy. Nat. Med. 29, 95–103. https://doi.org/10.1038/s41591-022-02045-1.

Rinaldi, M., Petrarca, M., Romano, A., Vasco, G., D'Anna, C., Bibbo, D., et al., 2020. Progression of muscular co-activation and gait variability in children with Duchenne muscular dystrophy: a 2-year follow-up study. Clin. Biomech. 78, 105101. https://doi.org/10.1016/j.clinbiomech.2020.105101.

Robertson, G., Caldwell, G., Hamill, J., Kamen, G., Whittlesey, S., 2013. Research methods in biomechanics, 2E. Human Kinetics. ISBN: 0-7360-8340-0. 440 p.

Romilly, D.P., Anglin, C., Gosine, R.G., Hershler, C., Raschke, S.U., 1994. A functional task analysis and motion simulation for the development of a powered upper-limb orthosis. IEEE Trans. Rehab. Eng. 2, 119–129. https://doi.org/10.1109/86.331561.

Rosen, J., Perry, J.C., Manning, N., Burns, S., Hannaford, B., 2005. The human arm kinematics and dynamics during daily activities - toward a 7 DOF upper limb powered exoskeleton. In: ICAR '05. Proceedings., 12th International Conference on Advanced Robotics, pp. 532–539. https://doi.org/10.1109/ICAR.2005.1507460.

Šenk, M., Chèze, L., 2010. A new method for motion capture of the scapula using an optoelectronic tracking device: a feasibility study. Comput. Methods Biomech. Biomed. Eng. 13, 397–401. https://doi.org/10.1080/10255840903263945.

Sobierajska-Rek, A., Jabłońska-Brudło, J., Dąbrowska, A., Wojnicz, W., Meyer-Szary, J., Wierzba, J., 2024. Timed rolling and rising tests in Duchenne muscular dystrophy ambulant boys: a feasibility study. Minerva Pediatr. 76. https://doi.org/10.23736/S2724-5276.21.05977-2.

Spörri, J., Schiefermüller, C., Müller, E., 2016. Collecting kinematic data on a ski track with optoelectronic Stereophotogrammetry: a methodological study assessing the feasibility of bringing the biomechanics lab to the field. PLoS One 11, e0161757. https://doi.org/10.1371/journal.pone.0161757.

Stegeman, D., Hermens, H., 2007. Standards for Suface Electromyography: The European Project Surface EMG for Non-invasive Assessment of Muscles (SENIAM), 1.

Summers, J., Larkin, D., Dewey, D., 2008. Activities of daily living in children with developmental coordination disorder: dressing, personal hygiene, and eating skills. Hum. Mov. Sci. 27, 215–229. https://doi.org/10.1016/j.humov.2008.02.002.

Szabo, S.M., Salhany, R.M., Deighton, A., Harwood, M., Mah, J., Gooch, K.L., 2021. The clinical course of Duchenne muscular dystrophy in the corticosteroid treatment era: a systematic literature review. Orphanet J. Rare Dis. 16, 237. https://doi.org/10.1186/s13023-021-01862-w.

The FDG Robotic Rehabilitation Group, Germanotta, M., Gower, V., Papadopoulou, D., Cruciani, A., Pecchioli, C., et al., 2020. Reliability, validity and discriminant ability of a robotic device for finger training in patients with subacute stroke. J. NeuroEng. Rehabil. 17, 1. https://doi.org/10.1186/s12984-019-0634-5.

Troka, M., Wojnicz, W., Szepietowska, K., Podlasiński, M., Walerzak, S., Walerzak, K., et al., 2022. Towards classification of patients based on surface EMG data of temporomandibular joint muscles using self-organising maps. Biomed. Signal Process. Control 72, 103322. https://doi.org/10.1016/j.bspc.2021.103322.

van der Kruk, E., Reijne, M.M., 2018. Accuracy of human motion capture systems for sport applications; state-of-the-art review. Eur. J. Sport Sci. 18, 806–819. https://doi.org/10.1080/17461391.2018.1463397.

Voet, N.B.M., Saris, C.G.J., Thijssen, D.H.J., Bastiaans, V., Sluijs, D.E., Janssen, M.M.H.P., 2022. Surface electromyography thresholds as a measure for performance fatigability during incremental cycling in patients with neuromuscular disorders. Front. Physiol. 13, 821584. https://doi.org/10.3389/fphys.2022.821584.

Wernio, E., Wasilewska, E., Czaja-Stolc, S., Śledzińska, K., Wierzba, J., Szlagatys-Sidorkiewicz, A., et al., 2024. Nutritional issues among children with Duchenne muscular dystrophy—incidence of deficiency and excess body mass. Nutrients 16, 2143. https://doi.org/10.3390/nu16132143.

Winter, D.A., 2009. Biomechanics and Motor Control of Human Movement, 4th ed. Wiley, Hoboken, N.J.

Wojnicz, W., Sobierajska-Rek, A., Zagrodny, B., Ludwicki, M., Jabłońska-Brudło, J., Forysiak, K., 2022. A new approach to assess quality of motion in functional task of upper limb in Duchenne muscular dystrophy. Appl. Sci. 12, 12247. https://doi.org/10.3390/app122312247.

Wood, C.L., Straub, V., Guglieri, M., Bushby, K., Cheetham, T., 2016. Short stature and pubertal delay in Duchenne muscular dystrophy. Arch. Dis. Child. 101, 101–106. https://doi.org/10.1136/archdischild-2015-308654.

Yoon, J.A., Park, H.E., Kim, J., Son, J., Shin, Y.B., 2022. Corticosteroid use and bone health management for Duchenne muscular dystrophy in South Korea. Sci. Rep. 12, 11300. https://doi.org/10.1038/s41598-022-15510-1.

Yu, B., Gabriel, D., Noble, L., An, K.-N., 1999. Estimate of the Optimum Cutoff Frequency for the Butterworth Low-Pass Digital Filter. https://doi.org/10.1123/jab.15.3.318.