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Gait deviations in Duchenne muscular dystrophy – part 1.

A systematic review

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Highlights.

- We conducted a systematic literature review to map gait deviations in DMD
- Only nine articles analyzed gait deviations in children with DMD
- Generalizability of these nine papers is poor
- Chances of a type I error are high in six of the nine included articles
- There is need for standardized measurement- and analyses methods

Abstract.

Background.

Although prolonged ambulation is considered important in children with Duchenne muscular dystrophy (DMD), articles describing gait deviations in DMD are scarce.

Research question.

Therefore, our research questions were the following: 1) what are the most consistently reported spatiotemporal-, kinematic-, kinetic-, and muscle activity deviations in children with DMD in literature, 2) what is the quality of the studies describing these deviations, and 3) is there need for further research?

Methods.

We conducted a systematic literature search for studies published before the end of June 2017 in six online databases. We created a data extraction form to define information on materials and methods and

on the analyzed gait parameters for each paper included in the review. If enough information was available, we calculated standardized mean differences (SMDs).

Results.

The search yielded nine articles, but generalizability was poor. Seventy-nine parameters were analyzed by seven research groups, but they only agreed on a decrease in walking speed (minimal SMD: 1.26), stride length (1.83), step length (1.80), dorsiflexion during swing (1.43), maximal power generation at the hip (0.92), maximal knee extension torque (0.99), maximal dorsiflexion torque (-1.30), and maximal power generation at the ankle (0.92), and an increased knee range of motion (-0.82) in DMD.

Significance.

In order to keep children with DMD ambulant as long as possible, a clear understanding of their pathological gait pattern is necessary. However, gait deviations in DMD appear not well defined. Previous studies appear to be of an exploratory nature while using predefined gait parameters to assess an undirected null hypothesis. This made them prone to regional focus bias, thereby increasing the chance of a type I error. Therefore, further research is required to define the altered gait pattern in children with DMD.

Keywords: Duchenne muscular dystrophy, 3D gait analysis, gait deviations, muscle weakness, neuromuscular disease

Introduction.

Duchenne muscular dystrophy (DMD) is the most common of muscular dystrophies affecting one in 3500-5000 boys that are born [1]. DMD is caused by a defective gene on the X-chromosome, which codes for the protein dystrophin. Dystrophin is expected to play an important role in “the stability of the muscle cell membrane in protecting the muscle fibers from contraction induced damage” [2]. When damage finally gets the overhand, muscles will predominantly consist of fibrofatty tissue [1]. So far, no cure has been found, and the affected children usually die in their third or fourth decade due to cardiac failure or pulmonary infections [1,3]. One of the treatment goals in children with DMD is to keep them ambulant as long as possible, aiming to preserve a clinically important function and to postpone spinal deformities and muscle contractures [1,4]. To achieve this goal, the altered gait pattern of children with DMD needs to be delineated and the potential underlying causes of their gait deviations need to be specified.

While 3D gait analysis to assess walking performance is part of the standard clinical care in children with neuromotor problems, such as cerebral palsy [5,6], this evaluation procedure is less often applied in children with DMD. In a clinical setting, walking ability in DMD is generally evaluated by means of the 6-minute walk test [7], which is also used to assess treatment effect in clinical trials [8]. Although the 6-minute walk test has been found to be a valid general measurement tool [7,8], it does not allow for a detailed analysis of joint kinematics, -kinetics, or muscle activity patterns of gait.

In 1981, Sutherland et al. were one of the first researchers using motion analysis to objectively quantify gait deviations in children with DMD [4]. They determined that three kinematic features could quantify disease progression: increased pelvic tilt, decreased dorsiflexion angle during swing and decreased cadence [4]. More recent studies used 3D motion analysis systems to study the gait pattern in children with DMD thereby calculating several spatiotemporal-, kinematic-, kinetic-, and/or muscle activity parameters [9–16]. However, their results are difficult to compare, due to differences in measurement

methods, data analysis procedures and extracted parameter sets [9–15,17]. Hence, even though prolonged ambulation is considered important in children with DMD [1], their gait deviations appear not well-defined.

Therefore, the goals of part 1 of this research project were to create an overview of published studies that applied motion analysis to objectively quantify gait deviations in children with DMD, to provide a summary of the alterations in spatiotemporal-, kinematic-, kinetic- and muscle activity parameters reported in literature, and to assess the quality of the included studies. To meet this goal, we conducted a systematic literature search for studies published before the end of June 2017, based on exploration of six online databases.

Material and methods.

Literature search.

First, the PICO (Population, Intervention, Comparison and Outcome) tool was applied to assist with forming relevant keywords to conduct the systematic search [18]. Based on this PICO, the following medical subheadings (MeSH) were chosen: “muscular dystrophy, Duchenne” and “gait”. If usage of MeSH-terms was not allowed, the following keywords were used: “Duchenne” or “Duchenne’s”, combined with “dystrophy” or “muscular dystrophy” or “disease”. Also, only the abbreviation “DMD” was applied. As a replacement for the MeSH-term “gait”, we used the keywords: “gait”, “gait analysis” or “3D gait analysis”. One reviewer (MG) checked four databases (Campell, Cochrane, Dare, and PubMed) to verify whether there were already previous published systematic reviews summarizing gait deviations in children with DMD. All systematic reviews published before June 2017, written in English, and using objective motion analysis techniques to study gait in children with DMD were included. We thereby defined ‘objective

motion analysis techniques', as measurement methods using 3D motion analysis systems from which spatiotemporal-, kinematic-, kinetic-, and/or muscle activity data could be derived. Titles and abstracts of the systematic reviews were checked by one reviewer (MG) to verify if they were relevant.

In the search for relevant articles, the same MeSH-terms and keywords were used as during the search for systematic reviews. Further, the following inclusion criteria were defined: 1) study population had to be aged between five and 18 years old, 2) a group of TD children needed to be used as controls, and 3) gait parameters needed to be objectively quantified. Studies only describing treatment outcomes, animal studies, or conference abstracts/-proceedings were excluded.

Then, three reviewers, two junior researchers (JD and LV) and one senior researcher (MG) systematically searched six electronic databases (PubMed, Web of Science, MedLine, Embase, Cochrane, and Google Scholar), to find articles objectively describing gait deviations in children with DMD. Titles and abstracts were screened by each reviewer individually (MG, JD, and LV). Eligibility of the full text papers was checked by four reviewers (MG, MVH, JD, and LV). In case of a difference in opinion between the reviewers (MG, MVH, JD, and LV) whether a study met the inclusion criteria, a consensus meeting was held with the option of consulting an additional senior researcher (KD). Finally, the reference lists of the included papers were also checked by one reviewer (MG). When a paper from the reference lists was considered eligible (based on title and abstract), the same procedure was followed as for the articles found in the online databases. If studies were not available online or in the library of the KU Leuven, a request was sent to the authors. A maximal waiting period of six weeks after the last search was employed.

Data extraction.

From the articles that were included in the review, information was extracted with the help of a custom-made data extraction form, based on the methods and results sections of the STROBE (Strengthening the

Reporting of OBservational studies in Epidemiology) checklist for case-control studies [19,20]. Two reviewers (MG and MVH) filled in the data extraction form individually and compared their results. In case of differences between the two reviewers, the respective paper was checked. If the differences could not be solved, an additional reviewer (KD) was consulted to reach consensus.

Data extracted from each paper consisted of a description of the study populations, measurement systems, measurement procedures, applied statistics, and selected gait parameters. With respect to study population, we extracted sample size, including the rationale behind the sample size, such as indicating whether a power analysis was conducted prior to the data collection. Information on how the diagnosis of DMD was reached was also obtained. Additionally, the reviewers (MG and MVH) summarized age, anthropometrics, functional level (Vignos-scale), and steroid regiment for the children with DMD, since this information was considered to be important for the generalizability of the study results [12,21–23]. Specifications of the gait analysis procedure, including information on measurement devices, sample frequency, filter procedures, and marker protocols were also extracted. Especially different marker configurations could give different outcomes with respect to gait kinematics [24].

Concerning the gait analysis measurements, the same two reviewers summarized information about walking speed (self-selected or faster/slower) [25], as well as whether the children walked barefoot or with shoes (with or without orthoses), and the number of gait cycles that were included in the analyses. We also considered it important to check how many parameters were analyzed, what type of statistics was applied, and which critical p-value was used in each study.

Then, spatiotemporal-, kinematic-, kinetic-, and muscle activity parameters, that were reported to be significantly different between DMD and TD, were summarized. For all parameters, we extracted the mean and standard deviation. When information about the mean and/or standard deviation was missing, we tried to estimate the required values via inspection of available figures and/or we used equations one

and two [26]. These formulas allow for conversion of the median (m) and the minimal (a) and maximal (b) values into mean (μ) values and standard deviations (σ) [26]. In equation one, N is the sample size.

$$1) \quad \mu = \frac{a+2*m+b}{4} + \frac{a-2*m+b}{4*N}$$

$$2) \quad \sigma = \frac{b-a}{4}$$

For all parameters, outcome units were checked, since normalization to bodyweight (in case of kinetics) or leg length (or height) (in case of several spatiotemporal parameters) could have an influence on the outcomes [27]. If possible, the outcomes of the different studies were converted to the same units to allow for better comparison.

Data analysis.

In case of sufficient information, we calculated the standardized mean difference (SMD) (equation three), including its confidence intervals (equation four) [28,29]. For equation three, μ_{td} is the mean value of the assessed parameter of the TD children and μ_{dmd} the mean value of the children with DMD for that same parameter. σ_{td} and σ_{dmd} are the standard deviations of both groups. The SMD represents the effect size. While the p-value indicates whether two groups significantly differ from each other for the assessed parameter, the effect size indicates how large this difference is [30].

$$3) \quad SMD = \frac{\mu_{td} - \mu_{dmd}}{\sqrt{(\sigma_{td}^2 + \sigma_{dmd}^2)/2}}$$

In equation four, 95% confidence intervals (*CI*) of the SMD are determined with the standard error (*SE*). The latter was calculated with equation five, in which N_{td} is the sample size of the TD children and N_{dmd} the sample size of the children with DMD [31].

$$4) \quad CI = SMD \pm 1.96 * SE$$

$$5) \quad SE = \sqrt{\frac{N_{td} + N_{dmd}}{N_{td} * N_{dmd}} + \frac{SMD^2}{2 * (N_{td} + N_{dmd})}}$$

A parameter was considered altered in children with DMD in the following instances: 1) at least two research groups should have found the same outcomes both with a SMD of 0.80 or higher, or 2) when SMDs could not be calculated due to insufficient data, at least two research groups should have similar results and an additional research group should report a SMD of 0.80 or higher for that same parameter. For both requirements, the CI of the effect sizes should not include zero, and no contradictory results were allowed to be reported by the other studies. When a research group reported no significant differences, their results were not taken into consideration, unless there was an effect size of 0.80 or higher. Finally, we only compared the results of gait parameters while walking at self-selected speed.

Results.

Literature search.

No systematic reviews, describing gait deviations in children with DMD, were found in the online databases. Based on title and abstract and after removal of duplicates, the systematic search yielded 16 articles [4,9–17,21,32–36]. All 16 articles could be obtained via the online databases or through the library

of the KU Leuven. Of these 16 articles, seven articles were excluded. Reasons for exclusion were: one article appeared to be a conference abstract [34], one research group used a gait classification tool instead of extracted gait parameters [15], two studies did not use a motion analysis system (as defined in our inclusion criteria) to analyze gait in DMD [17,36], and in three studies a control group of TD children was missing [14,21,33]. A total of nine articles were included in the review [4,9–13,16,32,35]. Reference lists of these nine articles were reviewed, which led to one potentially relevant paper based on title and abstract [37]. However, after reading the full-text, the paper was excluded by all four reviewers, because it was a narrative literature review, without a systematic literature search. A full overview of the results of the systematic search is visualized in Figure 1.

Data extraction.

The results of the data extraction can be found in Tables 1 to 5. Information about the study population, measurement systems, measurements, and statistical analysis can be found in Table 1. Only one research group completely described the study population [12]. In all other articles, information about the control group [4,16,32,35], and/or anthropometrics or treatment regimen of the children with DMD was missing [4,9–11,13,16,32,35]. Further, in none of the articles a rationale for the sample size was given [4,10–13,16,32,35]. Based on the extracted subject information, the same study population appeared to be used in two of the three papers published by the same research group [10,11]. In the study of Doglio et al. (2011) there was an effect for age, height, and weight indicating that the TD children were older, taller, and heavier than the children with DMD (SMD = 1.43 (0.53 – 2.32), 1.88 (0.96 – 2.79) and 1.47 (0.58 – 2.37) respectively) [13]. In another research group, the TD children were also taller than the children with DMD (SMD = 1.69 (0.82 – 2.56)) [10,11].

Measurement systems were described, but the authors did not always report sample frequency or filtering procedures. Marker protocols were reported in all papers [4,9–13,16,32,35]. One research group failed to mention whether the children walked barefoot or with shoes [32], two research groups did not report at what walking speed the gait analysis was performed [32,35], and four research groups did not sufficiently define the number of gait cycles included in the analyses [9,11,16,32,35]. Statistics were clearly reported, but a rationale behind the parameter selection was not always given [11–13,32].

Concerning the extracted gait parameters, a total of 79 parameters were reported to be altered, of which eight were spatiotemporal- (Table 2), 22 kinematic- (Table 3), 19 kinetic- (Table 4), and 30 muscle activity (Table 5) parameters. All parameters could be converted to the same outcome units with exception of the muscle activity parameters in the studies of Sutherland et al. (1981) and Ropars et al. (2016), and knee-extension torque in the study of Boccardi et al. (1997) [4,16,32].

In all articles but two, spatiotemporal parameters were analyzed [9,35]. Two research groups normalized several of the spatiotemporal parameters to height [12,32]. Another study took the effect of walking speed on gait kinematics and -kinetics into consideration by comparing the gait parameters of the children with DMD with gait parameters of TD children that walked slower (v_0) than their natural walking speed (v_1) [11]. For non-normalized self-selected walking speed, all studies agreed that children with DMD walked slower than TD children with SMDs ranging from 1.26 to 3.20 [4,10–13,16]. However, this difference disappeared in the study of D'Angelo et al. (2009) when walking speed was normalized to height (SMD = 0.67 (-0.10 – 1.44) [12]. Additionally, a decrease in stride and step length was reported in the children with DMD with a minimal SMD of 1.71 and 1.80, respectively [12,13,16,32].

Mainly sagittal kinematic- and kinetic parameters were analyzed (34 of the 41 parameters). Only three research groups reported significantly different features in the children with DMD in the coronal or transversal plane [4,12,13]. Five of the seven research groups described significant differences in a total of 22 kinematic gait parameters [4,11–13,32], but for only two of these parameters their findings were

considered consistent: 1) an increased total range of motion of the knee [12,13], and 2) a decreased dorsiflexion angle in swing [4,11–13] in children with DMD when compared to the TD children (Table 3). For these parameters minimal SMDs were -0.82 and 1.43 respectively.

Five research groups reported changes in 19 kinetic gait parameters (Table 4) [9–13,32]. Four of these parameters were consistently reported to be decreased in children with DMD when compared to the control group: 1) maximal power generation at the hip at H3 (as described by Winter) (minimal SMD = 0.92) [11,12,38], 2) maximal net knee extension torque (minimal SMD = 0.99) [11,13,32,35], 3) maximal dorsiflexion net joint torque (minimal SMD = -1.30) [10,11,13], and 4) maximal power generation at the ankle during push off (minimal SMD = 0.92) [11–13].

Finally, three research groups analyzed muscle activity patterns in children with DMD [4,16,32], but only two compared the muscle activity data with data of their control group [4,16] (Table 5). No consistent changes in muscle activity data could be reported, since SMDs could only be calculated for one study [16].

Discussion.

The aim of this systematic review was to summarize the gait deviations in children with DMD reported in literature, to assess the quality of the included studies, and to analyze if there is need for further research. Our systematic literature search yielded nine eligible articles of which three were of the same research group. Generalizability of the results reported in these nine studies was poor, due to an insufficient description of the study population and measurement devices. Also, a rationale for the sample size or for the selected parameters was often not given.

Furthermore, a total of 79 gait parameters were extracted and analyzed of which only 18 parameters were reported by three or more research groups, suggesting that most studies were of an exploratory nature. However, by using predetermined gait parameters to answer an undirected hypothesis, the results were

prone to regional focus bias, thereby increasing the chance of a type I error [39,40]. Finally, by extracting kinematic- and kinetic parameters from the individual joints, these parameters are considered as independent variables, which is not correct. For example, in typical gait, activation of the soleus muscle in stance influences ankle kinematics and -kinetics directly, but also indirectly affects the knee- and hip joints [41]. When this dynamic coupling is not taken into consideration, it will yet cause another form of bias, i.e. the inter-component covariance bias [39]. One way to take both biases into consideration, is by using an appropriate analysis methods to answer an undirected hypothesis, such as statistical parametric mapping (SPM) [42]. This technique will be used to analyze gait deviations in children with DMD in part 2 of this research on gait in DMD.

In the seven papers analyzing spatiotemporal parameters in children with DMD, three of the eight parameters were considered decreased in the children with DMD (walking speed, stride-, and step length) [4,10–13,16,32]. It is well-known that spatiotemporal parameters are related to a subject's anthropometry [27,43]. Therefore, it is recommended to normalize the spatiotemporal parameters accordingly [27,43]. Due to the long-term use of corticosteroids, children with DMD are characterized by a short stature and increased bodyweight when compared to age-matched TD children [3]. However, for the studies included in this review, this characterization mainly applies to height [10,11,13]. The differences in spatiotemporal parameters between DMD and TD were found for non-normalized values and the lack of normalization might have influenced the outcomes. Yet, D'Angelo et al (2009) normalized walking speed and stride length to height, and found that for walking speed the significant differences between the two groups disappeared after normalization, while they remained present for stride length [12].

Normalization of gait kinetics to bodyweight is also recommended [44,45], and has been performed by the three research groups analyzing gait kinetics [11–13]. In two studies of the same research groups, net

joint torques were not normalized [9,10], which could have an effect on the results and conclusions that were made. However, this effect is very small, since the SMD (CI) for weight was 0.34 (-0.47 – 1.14).

When looking at the changes in kinematic and kinetic parameters, the children with DMD showed an increased total range of motion at the knee [12,13], a decreased dorsiflexion during swing [4,11–13], a decreased maximal power generation at the hip at the end of stance (representing the H3-burst as described by Winter) [11,12,38] and ankle before push-off (the A2-burst as described by Winter) [11–13,38], and a decreased maximal net knee extension torque and dorsiflexion torque during stance [9,11,13,32]. Muscle weakness of the dorsiflexors and/or muscle contractures of the plantar flexors, potentially cause a decrease in dorsiflexion angle during stance and swing [1,10,46]. However, during stance the results reported by the included studies were not consistent [4,10,12,13,32]. A possible explanation for these discrepancies could be the presence of a midfoot break in children with DMD, which would explain the slightly higher dorsiflexion angle during stance in two of the included studies [10,32]. This foot deformity is translated in an overestimation of ankle dorsiflexion, due to the lack of detailed foot models that would allow decomposed quantification of the hind- and midfoot [47,48]. By positioning the ankle more in plantar flexion during stance [4,12,13], the ground reaction force can be positioned closer to or in front of the knee joint center, thereby reducing internal net knee extension torque accommodating the weak quadriceps. However, this can force the knee in hyper-extension, partially explaining the increase in total range of motion of the knee [12,13]. Further, due to the decreased dorsiflexion angle during stance, the angular velocity needed to generate enough power for push-off at the ankle is limited, explaining the lower maximal ankle power generation. Additionally, an increase in knee flexion is required to avoid tripping or falling due to the decreased dorsiflexion angle during swing, further contributing to the increase in total knee range of motion [12,13]. It has previously been suggested that the lower power generation at the hip could be explained by weakness of the hip extensors, resulting in a pelvic anterior tilt [11,12,32], and consequently a more flexed position of the hip [4,11,32]. This could

result in a less optimal position for the hip flexors to generate enough power for pull off [38]. Also, muscle contractures and weakness of the hip flexors could have contributed to this decrease in power at the hip joint [1,34]. Finally, the lower dorsiflexion torque at the beginning of stance can be explained by the absence of a heel-strike, whereby the children with DMD either place the foot in plantar flexed position or flat on the ground. However, all these explanations assume that muscle weakness is the main contributor to these alterations, and although this reasoning is sound, it has not yet been verified.

Similar to spatiotemporal- and kinetic parameters, muscle activity data is also subjective to processing factors [49]. The values needed to calculate SMDs and information about data processing of electromyographic (EMG) data is missing in the study of Sutherland et al. (1981) [4], which makes it difficult to compare their results with the outcomes of Ropars et al. (2016) [16]. However, in general, muscle activity seems to be increased in children with DMD. Boccardi et al. (1997) described muscle activity patterns of the trunk, but they did not compare their results with the control group, so it remains unsure whether muscle activity was altered in their group of children with DMD [32].

Finally, with exception of the study of Doglio et al. (2011) [13], none of the studies included in the review accounted for the progressive nature of the disease, which was evident in the wide age range of the study populations [4,9–12,16,32,35]. This may also explain the differences in altered gait parameters between studies.

One of the limitations of this review was that we only included articles that were written in English. Based on this inclusion criterium, we rejected two papers, of Patte et al. (2000) [50] and Ando et al. (1992) [51], (written in French and Japanese, respectively), making the review prone to language bias. However, while inclusion of those two studies might have added additional gait parameters to the already extensive list, we expect that the overall conclusions would remain the same [52]. Further, English is the language in which almost 80% of research is published [53], thus we are confident that we covered most of articles published on this topic.

Conclusion.

Although sustained walking ability is considered important in DMD, there is limited agreement on how DMD gait deviates from a typical gait pattern concerning spatiotemporal-, kinematic-, kinetic-, and muscle activity parameters. Due to the discrepancies between the nine included studies there remains a substantial lack of generalizability of the outcomes. Therefore, for future studies analyzing gait deviations in children with DMD, it is important that the materials and methods (including statistics) are well described. Also, since 3D gait analysis in children with DMD appears to be in an exploratory phase, it might be more appropriate to start mapping gait deviations in this population by utilizing an undirected null hypothesis, instead of performing analyses of predetermined gait parameters.

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Conflicts of interest.

There were no conflicts of interest.

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Figure 1. Search strategy and results reviews and articles included in the review.
Abbreviations: DMD = Duchenne Muscular Dystrophy; N = number; TD = Typical developing ;
WOS = Web Of Science

Table 1. The results of the data extraction concerning study population, measurement system, measurements and statistics are summarized below.

If available, mean values and standard deviations (SD) of the extracted parameter are given. In each cell, first information about the children with DMD is given (mean $\pm$ SD), then for the TD children (mean $\pm$ SD) and finally the effect size (in cursive). Effect sizes of 0.80 or higher are printed in bold. If information about the mean or SD was missing, the cell was left empty. If effect sizes could not be calculated or estimated from figures, this is indicated as *insufficient data*.

	Sutherland et al 1981	Khodadadeh et al 1986	Boccardi et al 1997	Gaudreault et al 2006	Gaudreault et al 2009	Gaudreault et al 2010	D'Angelo et al 2009	Doglio et al 2011	Ropars et al 2016
Study population									
Sample size	21 DMD ¹ 15 TD	7 DMD 21 TD	6 DMD 15 TD	1 DMD 1 TD	11 DMD 14 TD	11 DMD 14 TD	21 DMD ² 10 TD	15 DMD 9 TD	16 DMD 15 TD
Rationale sample size				N.A.					
Diagnosis DMD confirmed	Yes				Yes	Yes	Yes	Yes	Yes
Gender	21 Males	7 Males 21 Males		1 Male	11 Males	11 Males	21 Males 10 Males	15 Males 9 Males	16 Males 15 Males
Age (years)	7.5 $\pm$ 1.6 ³ 7.0	9.0 $\pm$ 2.0 9.0 $\pm$ 1.8 <i>0.00 (-0.86 – 0.86)</i>	9 $\pm$ 3	10.6 11.4	9.2 $\pm$ 2.6 9.7 $\pm$ 1.9 <i>0.22 (-0.58 – 1.02)</i>	9.3 $\pm$ 2.6 ⁴ 9.5 $\pm$ 1.9 <i>0.09 (-0.71 – 0.88)</i>	6.8 $\pm$ 2.4 7.4 $\pm$ 1.2 <i>0.32 (-0.45 – 1.08)</i>	6.1 $\pm$ 0.7 7.5 $\pm$ 1.2 <i>1.43 (0.53 – 2.32)</i>	8.7 $\pm$ 2.0 9.4 $\pm$ 2.2 <i>0.33 (-0.39 – 1.05)</i>
Height (m)			1.30 $\pm$ 0.11	1.29 1.44	1.27 $\pm$ 0.06 1.42 $\pm$ 0.11 <i>1.69 (0.82 – 2.56)</i>	1.27 $\pm$ 0.06 1.42 $\pm$ 0.11 <i>1.69 (0.82 – 2.56)</i>	1.19 $\pm$ 0.13 1.27 $\pm$ 0.07 <i>0.77 (-0.02 – 1.55)</i>	1.14 $\pm$ 0.08 1.31 $\pm$ 0.10 <i>1.88 (0.96 – 2.79)</i>	

¹ Divided the DMDs into three groups with respect to disease progression: early, transitional (trans) and late. Data was collected longitudinally over 4.5 years, so some children might be present in all three groups (early, trans, and late)

² Treated and untreated (with steroids) boys with DMD were grouped together in this review for clarity

³ Mean and standard deviation were estimated with the equations described by Hozo et al. (2015).

⁴ There is a small discrepancy between the values in the table and the values in the text. We have used the values from table 1.

Weight (kg)			28 ± 10	25.2 36.4	30.2 ± 8.1 32.8 ± 7.4	30.2 ± 8.1 32.8 ± 7.4	24.1 ± 8.5 25.0 ± 4.3	20.6 ± 4.4 27.7 ± 5.2	
Vignos-scale				3	0.34 (-0.47 – 1.14) 5 ≤	0.34 (-0.47 – 1.14) 5 ≤	0.58 (-0.20 – 1.36)	1.47 (0.58 – 2.37)	3.4 ± 1.4
Steroids (type)						Yes (11/11)	Prednisone (4) Deflazacort (10)		
Steroid dosage (ml/kg/day)							Prednisone: 0.75 Deflazacort: 0.90		
Measurement system									
Camera's [sf]	Photo Sonics, Hycam, Bolex [15-24 Hz]	ORLAU []	ELITE [100 Hz]	Optotrak [60 Hz]	Optotrak [60 Hz]	Optotrak [60 Hz]	ELITE2002 [100 Hz]	Vicon [60 Hz]	Optoelectrical system or Vicon []
Force plates [sf]	Described []	Kistler []	Kistler []	AMTI [600 Hz]	AMTI [600 Hz]	AMTI [600 Hz]	Kistler []	Kistler []	N.A.
Marker model used	Described		Described	Described	Described	Described	Davis' protocol	Davis' protocol	Davis' protocol
EMG [sf]	Textronix oscilloscope, Soltec oscillography []	N.A.	Telemetric system	NORAXON Telemyo []	N.A.	N.A.	N.A.	N.A.	CHRU Brest, 16-channel [1084 Hz]
Description filters used	Yes			Yes	Yes	Yes			Yes (for EMG)
Measurements									
Barefoot or shoes	Barefoot	Barefoot		Shoes	Shoes	Shoes	Barefoot	Barefoot	Barefoot
Walking velocities	v1			v1	v1	v1+ v0 (TD)	v1	v1	v1
Bi- or unilateral	Bilateral	Bilateral		Unilateral right	Unilateral right	Unilateral right	Bilateral	Bilateral	Bilateral
Number of GC included	1 (left or right)	2 left + 2 right (minimally)			Average of 3		Average of 3 (left + right pooled)	Average of 5 ≤	
Statistics									

Number of gait parameters	15 (31)	1	10	2	30	29	47 + 12 curves	26	
Rationale parameter selection	Yes	Yes		Yes	Yes			Yes	
Parametric or else	Parametric + Descriptive	Descriptive	Descriptive	Descriptive	Parametric	Parametric + Descriptive	Parametric	Parametric	Non-parametric
Test for group differences	Two-tailed t-test				Student's t-test	Student's t-test	Student's t-test	Student's t-test	Mann-Whitney U
Correction for multiple testing		N.A.	N.A.	N.A.					

Abbreviations in alphabetic order: DMD = Duchenne muscular dystrophy; GC = gait cycle; kg = kilograms; m = meters; N.A. = not applicable; TD = typical developing; sf = sample frequency; v0 = slower than self-selected walking speed; v1 = self-selected walking speed

Table 2. The results of the data extraction concerning the spatiotemporal parameters are summarized below. If available, mean values and standard deviations (SD) of the extracted parameter are given. In each cell, first information about the children with DMD is given (mean $\pm$ SD), then for the TD children (mean $\pm$ SD) and finally the effect size (in cursive). Effect sizes of 0.80 or higher are printed in bold. If information about the mean or SD was missing, the cell was left empty. If effect sizes could not be calculated or estimated from figures, this is indicated as *insufficient data*.

	Sutherland et al 1981	Khodadadeh et al 1986	Boccardi et al 1997	Gaudreault et al 2006	Gaudreault et al 2009	Gaudreault et al 2010	D'Angelo et al 2009	Doglio et al 2011	Ropars et al 2016
Spatiotemporal									
Walking speed [m/s]	DMD all ↓ 0.80 $\pm$ 0.34 1.14 $\pm$ 0.17 1.26 (0.54 – 1.99)		DMD ↓ 0.77 1.11		DMD ↓ 0.62 $\pm$ 0.12 1.02 $\pm$ 0.13 3.20 (2.01 – 4.38)	N.S. v0 DMD ↓ v1 0.62 $\pm$ 0.12 0.65 $\pm$ 0.09 1.02 $\pm$ 0.13 0.28 (-0.51 – 1.08) v0 3.20 (2.01 – 4.38) v1	N.S. ^N 0.99 $\pm$ 0.17 1.19 $\pm$ 0.17 0.67 (-0.10 – 1.44) ^N 1.35 (0.53 – 2.18)	DMD ↓ N.S. 1.06 $\pm$ 0.17 1.07 $\pm$ 0.18 0.06 (-0.77 – 0.88)	DMD ↓ 0.78 $\pm$ 0.18 1.21 $\pm$ 0.13 2.74 (1.76 – 3.72)
Cadence [steps/min]	DMD all ↓ 116 $\pm$ 24 144 $\pm$ 15 1.38 (0.65 – 2.12)				N.S. 100.4 $\pm$ 8.4 102.7 $\pm$ 13.6 0.20 (-0.59 – 1.00)	DMD ↑ v0 N.S. v1 100.4 $\pm$ 8.4 78.1 $\pm$ 9.7 102.7 $\pm$ 13.6 -2.46 (-3.50 – -1.41) v0 0.20 (-0.59 – 1.00) v1	N.S. 132 $\pm$ 20 131 $\pm$ 11 -0.06 (-0.82 – 0.69)	DMD ↑ 145.7 $\pm$ 14.9 121.2 $\pm$ 16.2 -1.57 (-2.51 – -0.64)	DMD ↓ 116.26 $\pm$ 16.8 129.68 $\pm$ 8.41 1.01 (0.26 – 1.76)
Stride length [m]	N.S. <i>Insufficient data</i>		DMD ↓ DMD ↓ ^N 0.72 0.58 ^N 0.86 0.65 ^N				DMD ↓ ^N 0.90 $\pm$ 0.06 1.04 $\pm$ 0.09 1.60 (0.75 – 2.45) ^N 1.83 (0.95 – 2.71)	DMD ↓ 0.87 $\pm$ 0.10 1.05 $\pm$ 0.11 1.71 (0.75 – 2.67)	DMD ↓ 0.81 $\pm$ 0.14 1.13 $\pm$ 0.12 2.45 (1.52 – 3.39)
Step length [m]	N.S. <i>Insufficient data</i>				DMD ↓ 0.37 $\pm$ 0.07 0.57 $\pm$ 0.06 3.07 (1.91 – 4.23)	DMD ↓ v0 DMD ↓ v1 0.37 $\pm$ 0.07 0.49 $\pm$ 0.03 0.57 $\pm$ 0.06 2.23 (1.23 – 3.23) v0 3.07 (1.91 – 4.23) v1		DMD ↓ 0.43 $\pm$ 0.05 0.52 $\pm$ 0.05 1.80 (0.83 – 2.77)	
Step width [m]							DMD ↑ ^N 0.09 $\pm$ 0.02		

^N Results for the normalized values

					0.06 ± 0.01 -1.88 (-2.77 – -1.00)		
Stride time [s]			DMD ↑		N.S. 0.94 ± 0.17 0.92 ± 0.07	DMD ↓ 0.84 ± 0.07 1.01 ± 0.16	
			<i>Insufficient data</i>		-0.10 (-0.85 – 0.65)	1.38 (0.46 – 2.29)	
Step time [s]	N.S.					DMD ↓ 0.42 ± 0.04 0.51 ± 0.08	
	<i>Insufficient data</i>					1.42 (0.50 – 2.34)	
Double support time during one GC [s]	N.S.		DMD ↑ 0.11 0.08			N.S. 0.18 ± 0.03 0.23 ± 2.81	
	<i>Insufficient data</i>		<i>Insufficient data</i>			1.67 (0.72 – 2.62)	

Abbreviations in alphabetic order: DMD = Duchenne muscular dystrophy; GC = gait cycle; m = meters; min = minutes; N.S. = not

significant; TD = typical developing; s = seconds; v0 = slower than self-selected walking speed; v1 = self-selected walking speed

Table 3. The results of the data extraction concerning gait kinematics are summarized below. If available, mean values and standard deviations of the extracted parameter are given. In each cell, first information about the children with DMD is given (mean $\pm$ SD), then for the TD children (mean $\pm$ SD) and finally the effect size (in cursive). Effect sizes of 0.80 or higher are printed in bold. If information about the mean or SD was missing, the cell was left empty. If effect sizes could not be calculated or estimated from figures, it is indicated as *insufficient data*.

	Sutherland et al 1981	Khodadadeh et al 1986	Boccardi et al 1997	Gaudreault et al 2006	Gaudreault et al 2009	Gaudreault et al 2010	D'Angelo et al 2009	Doglio et al 2011	Ropars et al 2016
Kinematics									
<u>Pelvis</u>									
<i>Sagittal</i>									
Total ROM [deg]								DMD $\uparrow$ 3.6 $\pm$ 1.1 2.0 $\pm$ 1.0 <i>-1.52 (-2.45 – -0.59)</i>	
Mean pelvic tilt [deg]			DMD $\uparrow$ <i>Insufficient data</i>				DMD $\uparrow$ 14.6 $\pm$ 5.0 10.0 $\pm$ 3.8 <i>-1.04 (-1.91 – -0.16)</i>		
Max pelvic tilt [deg]	DMD early $\uparrow$ <i>Insufficient data</i>							N.S. 13.5 $\pm$ 3.9 11.2 $\pm$ 3.2 <i>-0.64 (-1.49 – 0.20)</i>	
<i>Coronal</i>									
Downward movement double support [deg]			DMD $\uparrow$ <i>Insufficient data</i>					DMD $\uparrow$ ⁵ -3.0 $\pm$ 0.5 -1.3 $\pm$ 0.8 <i>2.55 (1.45 – 3.65)</i>	
<i>Transversal</i>									
Total ROM [deg]								DMD $\uparrow$ 18.4 $\pm$ 6.5	

⁵ Mean and standard deviation were estimated with the equations described by Hozo et al. (2015).

						13.3 ± 4.3 -0.93 (-1.79 – -0.06)	
External rotation stance [deg]						DMD ↑ ⁶ -9.2 ± 3.5 -6.5 ± 1.8 0.97 (0.10 – 1.84)	
<u>Hip</u>							
<i>Sagittal</i>							
Total ROM [deg]					N.S. 48.1 ± 5.6 47.0 ± 3.8 -0.23 (-0.99 – 0.53)	DMD ↑ 50.5 ± 6.8 45.2 ± 4.0 -0.95 (-1.82 – -0.08)	
Hip extension during H2 [deg]	DMD trans + late ↓ <i>Insufficient data</i>		DMD ↓ <i>Insufficient data</i>		DMD↓v0 DMD↓v1 -12.6 ± 11.7 -21.6 ± 7.5 -20.6 ± 8.6 -0.92 (-1.75 – -0.09) v0 -0.78 (-1.60 – 0.04) v1	N.S. -11.9 ± 4.7 -9.8 ± 3.6 0.50 (-0.34 – 1.34)	
Hip flexion during swing (H3) [deg]	DMD all ↑ <i>Insufficient data</i>		DMD ↑ <i>Insufficient data</i>		DMD↑v0 N.S. v1 20.3 ± 10.3 9.6 ± 9.3 18.3 ± 9.2 -1.09 (-1.94 – -0.24) v0 -0.20 (-1.00 – 0.59) v1	N.S. 38.5 ± 5.2 35.4 ± 5.8 -0.56 (-1.40 – 0.28)	
<i>Coronal</i>							
Mean hip ab-adduction during stance					DMD↓v0 DMD↓v1 2.2 ± 2.3 6.7 ± 1.8 5.9 ± 2.0 2.18 (1.18 – 3.17) v0 1.72 (0.79 – 2.64) v1	N.S. -2.9 ± 3.3 -1.6 ± 2.9 0.42 (-0.34 – 1.18)	
<u>Knee</u>							
<i>Sagittal</i>							
Total ROM [deg]					DMD ↑ 62.2 ± 6.0 58.2 ± 3.4 -0.82 (-1.60 – -0.04)	DMD ↑ 63.7 ± 7.2 56.8 ± 3.9 -1.19 (-2.08 – -0.30)	

⁶ Mean and standard deviation were estimated with the equations described by Hozo et al. (2015).

Knee flexion at initial contact [deg]			DMD ↑		N.S. 7.7 ± 5.9 8.9 ± 4.8		
			<i>Insufficient data</i>		<i>0.22 (-0.53 – 0.98)</i>		
Max knee flexion in stance [deg]					DMD ↓ 9.3 ± 6.7 16.1 ± 5.5		
Knee flexion in swing [deg]			DMD ↑	N.S. v0 N.S. v1 62.8 ± 5.1 58.5 ± 5.9 64.5 ± 6.5	DMD ↑ 60.6 ± 8.1 59.6 ± 5.4	DMD ↑ 64.8 ± 4.8 62.3 ± 4.4	
			<i>Insufficient data</i>	<i>-0.78 (-1.60 – 0.04) v0 0.29 (-0.50 – 1.08) v1</i>	<i>-0.15 (-0.90 – 0.61)</i>	<i>-0.54 (-1.38 – 0.30)</i>	
Max knee extension stance [deg]			DMD ↓		DMD ↑ -0.9 ± 5.8 2.9 ± 4.9	DMD ↑ 1.0 ± 4.5 5.4 ± 4.0	
			<i>Insufficient data</i>		<i>0.71 (-0.07 – 1.48)</i>	1.03 (0.16 – 1.91)	
Mean hyper-extension stance [% GC]					DMD ↑ 15.7 ± 14.5 4.9 ± 6.5		
					-0.96 (-1.75 – -0.17)		
<u>Ankle</u>							
<i>Sagittal</i>							
Total ROM [deg]					N.S. 26.8 ± 6.9 28.2 ± 5.7	DMD ↓ 25.2 ± 3.9 30.7 ± 5.2	
					<i>0.22 (-0.53 – 0.98)</i>	-1.20 (-2.09 – -0.30)	
Ankle angle at IC [deg]	DMD all ↑			N.S. v0 N.S. v1 -13.0 ± 8.5 -11.9 ± 5.8 -11.6 ± 5.5	DMD ↑ -4.6 ± 6.6 0.2 ± 3.6		
	<i>Insufficient data</i>			<i>0.15 (-0.64 – 0.94) v0 0.20 (-0.60 – 0.99) v1</i>	0.90 (0.12 – 1.96)		
Max plantar flexion angle [deg]						DMD ↓ -12.3 ± 6.4 -12.5 ± 5.7	
						<i>-0.03 (-0.86 – 0.79)</i>	
Max dorsiflexion angle stance [deg]	DMD late ↓		DMD ↑	N.S. 10.3 ± 6.2	DMD ↓ 10.6 ± 6.6	DMD ↓ 12.9 ± 4.9	

				9.9 ± 4.3		13.9 ± 2.2	18.3 ± 4.3	
	<i>Insufficient data</i>		<i>Insufficient data</i>	-0.07 (-2.85 – 2.70)		0.67 (-0.10 – 1.44)	1.17 (0.28 – 2.06)	
Max dorsiflexion angle swing [deg]	DMD all ↓			N.S. v0 N.S. v1 10.2 ± 6.2 9.9 ± 4.3 10.3 ± 6.1		DMD ↓ -5.0 ± 7.7 3.2 ± 2.6	DMD ↓ -1.9 ± 5.9 7.5 ± 2.2	
	<i>Insufficient data</i>			-0.06 (-0.85 – 0.73) v0 0.02 (-0.77 – 0.81) v1		1.43 (0.59 – 2.26)	2.11 (1.09 – 3.13)	
<i>Transversal</i>								
Foot progression angle [deg]	DMD early ↓ (ext) DMD trans + late ↑ (int)					N.S. -14.5 ± 7.9 -11.3 ± 4.9		
	<i>Insufficient data</i>					0.49 (-0.28 – 1.25)		

Abbreviations in alphabetic order: deg = degrees; DMD = Duchenne muscular dystrophy; ext = external rotation; GC = gait cycle; H2 = representing the H2-burst as described by Winter (1987); H3 = representing the H3-burst as described by Winter (1987); IC = initial contact; int = internal rotation; m = meters; N.S. = not significant; ROM = range of motion; TD = typical developing; v0 = slower than self-selected walking speed; v1 = self-selected walking speed

Table 4. The results of the data extraction concerning gait kinetics are summarized below. If available, mean values and standard deviations of the extracted parameter are given. In each cell, first information about the children with DMD is given (mean $\pm$ SD), then for the TD children (mean $\pm$ SD) and finally the effect size (in cursive). Effect sizes of 0.80 or higher are printed in bold. If information about the mean or SD was missing, the cell was left empty. If effect sizes could not be calculated or estimated from figures, it is indicated as *insufficient data*.

		Sutherland et al 1981	Khodadadeh et al 1986	Boccardi et al 1997	Gaudreault et al 2006	Gaudreault et al 2009	Gaudreault et al 2010	D'Angelo et al 2009	Doglio et al 2011
Kinetics									
<u>Hip</u>									
<i>Sagittal</i>									
Max hip extension torque at H1 [Nm/kg]			DMD ↓ <i>Insufficient data</i>			DMD ↓ v0 DMD ↓ v1 0.19 $\pm$ 0.11 0.33 $\pm$ 0.13 0.55 $\pm$ 0.16 1.16 (0.31 – 2.02) v0 2.62 (1.55 – 3.70) v1		N.S. 0.57 $\pm$ 0.21 0.58 $\pm$ 0.17 <i>0.05 (-0.77 – 0.88)</i>	
Max hip flexion torque at H2 [Nm/kg]				DMD ↑ -0.02 -0.01		N.S. v0 DMD ↓ v1 -0.38 $\pm$ 0.21 -0.36 $\pm$ 0.13 -0.53 $\pm$ 0.14 <i>0.11 (-0.68 – 0.90) v0</i> -0.84 (-1.66 – -0.02) v1		N.S. -0.60 $\pm$ 0.14 -0.69 $\pm$ 0.19 <i>-0.54 (-1.38 – 0.30)</i>	
Max power generation at H1 [W/kg]						N.S. v0 DMD ↓ v1 0.16 $\pm$ 0.11 0.26 $\pm$ 0.18 0.50 $\pm$ 0.28 <i>0.67 (-0.14 – 1.48) v0</i> 1.60 (0.69 – 2.50) v1		N.S. 0.86 $\pm$ 0.35 0.69 $\pm$ 0.30 <i>-0.52 (-1.36 – 0.32)</i>	
Max power generation at H3 [W/kg]						N.S. v0 DMD ↓ (v1) 0.22 $\pm$ 0.18 0.23 $\pm$ 0.12 0.48 $\pm$ 0.27 <i>0.07 (-0.72 – 0.86) v0</i> 1.13 (0.28 – 1.98) v1	DMD ↓ 0.45 $\pm$ 0.47 1.13 $\pm$ 0.94 0.92 (0.13 – 1.70)		
<i>Coronal</i>									

Mean abduction torque stance [Nm/kg]					DMD ↓v0 DMD ↓v1 ⁷ -0.30 ± 0.13 -0.59±0.20 -0.49±0.20 -1.72 (-2.64 – -0.80) v0 -1.13 (1.98 – -0.28) v1			
Power absorption mid-stance [W/kg]					DMD ↓v0 DMD ↓v1 ¹ 0.10 ± 0.07 0.01 ± 0.09 (v0 + v1) -1.12 (-1.96 – -0.27) v0 +v1			
<u>Knee</u>								
<i>Sagittal</i>								
Total torque ROM [Nm/kg]							DMD ↓ 0.47 ± 0.16 0.64 ± 0.20 0.94 (0.07 – 1.18)	
Max knee extension torque [Nm/kg]		DMD ↓ 10 (Nm) 60 (Nm) <i>Insufficient data</i>	DMD ↓ <i>Insufficient data</i>		DMD ↓v0 DMD ↓v1 ⁸ 0.07 ± 0.01 0.21±0.15 0.32±0.30 1.32 (0.45 – 2.19) v0 1.18 (0.32 – 2.03) v1		DMD ↓ 0.24 ± 0.13 0.39 ± 0.17 0.99 (0.12 – 1.86)	
Max knee extension torque at K3 [Nm/kg]					N.S. v0 DMD ↓ v1 0.05 ± 0.02 0.12±0.09 0.16±0.08 1.07 (0.23 – 1.92) v0 1.89 (0.94 – 2.83) v1			
Power absorption at K3 [W/kg]					N.S. v0 DMD ↓v1 -0.15 ± 0.18 -0.26±0.33 -0.49±0.32 -0.41 (-1.21 – 0.38) v0 -1.31 (-2.18 – -0.44) v1			
Power absorption at K4 [W/kg]					DMD ↓v0 N.S. v1 -0.31 ± 0.15 -0.13±0.07 -0.33±0.18 1.54 (0.64 – 2.44) v0			

⁷ Mean and standard deviation were estimated with the equations described by Hoze et al. (2015).

⁸ There appears to be a typo in table 2 of this paper, suggesting a higher knee extension torque in DMD instead of a lower.

						-0.12 (-0.91 – 0.67) v1		
<u>Ankle</u>								
<i>Sagittal</i>								
Total torque ROM [Nm/kg]							DMD ↓ 0.90 ± 0.12 1.35 ± 0.20 2.73 (1.60 – 3.86)	
Max dorsiflexion torque at A1 [Nm/kg]					DMD ↓ 0.08 -0.23 <i>Insufficient data</i>	DMD ↓ v0 DMD ↓ v1 0.04 ± 0.04 -0.16 ± 0.07 -0.23 ± 0.21 -3.51 (-4.76 – -2.26) v0 -1.79 (-2.72 – -0.85) v1	DMD ↓ -0.04 ± 0.03 -0.11 ± 0.07 -1.30 (-2.20 – -0.40)	
Plantar flexion torque at A2 [Nm/kg]				DMD ↓ 0.85 1.12 <i>Insufficient data</i>		N.S. v0 DMD ↓ v1 0.96 ± 0.90 1.07 ± 0.17 1.23 ± 0.15 <i>0.17 (-0.62 – 0.96) v0</i> <i>0.42 (-0.38 – 1.22) v1</i>	DMD ↓ 0.86 ± 0.11 1.24 ± 0.20 2.35 (1.29 – 3.42)	
Mean plantar flexion torque 5- 30% GC [Nm/kg]					DMD ↑ 0.25 ± 0.12 0.00 ± 0.12 -2.08 (-3.06 – -1.11)			
Mean plantar flexion torque 70- 90% GC [Nm/kg]					DMD ↓ 0.50 ± 0.21 0.69 ± 0.27 <i>0.79 (-0.03 – 1.60)</i>			
Total power ROM [W/kg]							DMD ↓ 2.88 ± 0.69 3.82 ± 1.45 0.83 (-0.03 – 1.69)	
Max power absorption at LR (A1) [W/kg]						N.S. v0 N.S. v1 -0.40 ± 0.10 -0.37 ± 0.17 -0.48 ± 0.11 <i>0.22 (-0.58 – 1.01) v0</i> <i>-0.76 (-1.58 – 0.06) v1</i>	N.S. -0.74 ± 0.22 -0.86 ± 0.39 <i>-0.38 (-1.21 – 0.45)</i>	
Max power generation at A2 [W/kg]						DMD ↓ v0 N.S. v1 1.36 ± 0.39 0.92 ± 0.42 1.70 ± 0.65	DMD ↓ 1.90 ± 0.75 2.71 ± 0.78	DMD ↓ 2.14 ± 0.58 2.95 ± 1.10

					-1.09 (-1.93 - -0.24) v0 0.63 (-0.17 - 1.44) v1	1.06 (0.26 - 1.86)	0.92 (0.05 - 1.79)	
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Abbreviations in alphabetic order: A1 = representing the A1-burst as described by Winter (1987); A2 = representing the A2-burst as described by Winter (1987); DMD = Duchenne muscular dystrophy; GC = gait cycle; H1 = representing the H1-burst as described by Winter (1987); H2 = representing the H2-burst as described by Winter (1987); H3 = representing the H3-burst as described by Winter (1987); IC = initial contact; Nm = Newton meters; Nm/kg = Newton meters per kilogram bodyweight; N.S. = not significant; ROM = range of motion; TD = typical developing; v0 = slower than self-selected walking speed; v1 = self-selected walking speed; W/kg = Watt per kilogram bodyweight;

Table 5. The results of the data extraction concerning muscle activity parameters are summarized below. If available, mean values and standard deviations of the extracted parameter are given. In each cell, first information about the children with DMD is given (mean $\pm$ SD), then for the TD children (mean $\pm$ SD) and finally the effect size (in cursive). Effect sizes of 0.80 or higher are printed in bold. If information about the mean or SD was missing, the cell was left empty. If effect sizes could not be calculated or estimated from figures, this is indicated as *insufficient data*.

	Sutherland et al 1981	Khodadadeh et al 1986	Boccardi et al 1997	Gaudreault et al 2006	Gaudreault et al 2009	Gaudreault et al 2010	D'Angelo et al 2009	Doglio et al 2011	Ropars et al 2016
Muscle activity									
Gluteus medius activity stance	DMD all $\downarrow$ <i>Insufficient data</i>								
Gluteus maximus activity swing	DMD trans + late $\uparrow$ <i>Insufficient data</i>								
Rectus femoris activity as % of max amplitude									DMD $\uparrow^9$ 0.24 $\pm$ 0.06 0.16 $\pm$ 0.07 -1.23 (-1.99 – -0.46)
Medial hamstrings activity as % of max amplitude									DMD $\uparrow^1$ 0.23 $\pm$ 0.08 0.18 $\pm$ 0.08 -0.63 (-1.35 – 0.10)
Medial hamstrings activity stance as % of max amplitude	DMD early + late $\downarrow$ <i>Insufficient data</i>								N.S. ¹ 0.26 $\pm$ 0.05 0.19 $\pm$ 0.03 -1.70 (-2.52 – -0.88)
Tibialis anterior activity stance as % of max amplitude	DMD late $\uparrow$ <i>Insufficient data</i>								DMD $\uparrow^1$ 0.25 $\pm$ 0.05 0.25 $\pm$ 0.12 0.00 (-0.70 – 0.70)

⁹ Mean and standard deviation were estimated with the equations described by Hozo et al. (2015).

Tibialis anterior activity 10-90% GC as % of max amplitude							DMD $\uparrow^1$ 0.28 $\pm$ 0.08 0.17 $\pm$ 0.06 -1.56 (-2.36 – -0.75)
Tibialis anterior activity 0-10% as % of max amplitude							DMD $\downarrow^1$ 0.34 $\pm$ 0.03 0.51 $\pm$ 0.09 2.53 (1.59 – 3.48)
Tibialis anterior activity 90-100% GC as % of max amplitude							DMD $\downarrow^1$ 0.32 $\pm$ 0.01 0.50 $\pm$ 0.08 3.16 (2.10 – 4.21)
Gastrocnemius activity 0-60% GC as % of max amplitude							Prolonged in DMD <i>Insufficient data</i>
Gastrocnemius activity 0-20% GC as % of max amplitude							DMD $\uparrow^1$ 0.27 $\pm$ 0.01 0.19 $\pm$ 0.03 -3.58 (-4.71 – -2.44)
KeR-EGI							DMD $\downarrow$ 76.88 $\pm$ 16.90 100.10 $\pm$ 7.90 1.76 (0.93 – 2.59)
EMG-PS global whole GC							DMD $\uparrow$ 0.17 $\pm$ 0.04 0.13 $\pm$ 0.02 -1.26 (-2.04 – -0.49)
EMG-PS rectus femoris whole GC							DMD $\uparrow$ 0.17 $\pm$ 0.06 0.13 $\pm$ 0.02 -0.89 (-1.63 – -0.16)
EMG-PS medial hamstrings whole GC							DMD $\uparrow$ 0.18 $\pm$ 0.04 0.15 $\pm$ 0.03 -0.85 (-1.58 – -0.11)
EMG-PS tibialis anterior							DMD $\uparrow$ 0.20 $\pm$ 0.03

whole GC							0.14 ± 0.04 -1.70 (-2.52 – -0.88)
EMG-PS gastrocnemius - soleus whole GC							DMD ↑ 0.17 ± 0.07 0.12 ± 0.03 -0.93 (-1.67 – -0.19)
EMG-PS global stance phase							DMD ↑ 0.18 ± 0.04 0.13 ± 0.01 -1.70 (-2.52 – -0.88)
EMG-PS rectus femoris stance phase							DMD ↑ 0.16 ± 0.06 0.11 ± 0.03 -1.05 (-1.81 – -0.30)
EMG-PS medial hamstrings Stance phase							DMD ↑ 0.18 ± 0.06 0.15 ± 0.03 -0.63 (-1.35 – 0.09)
EMG-PS tibialis anterior stance phase							DMD ↑ 0.18 ± 0.03 0.13 ± 0.02 -1.96 (-2.82 – -1.10)
EMG-PS gastrocnemius - soleus stance phase							DMD ↑ 0.20 ± 0.06 0.14 ± 0.03 -1.26 (-2.04 – -0.49)
EMG-PS global swing phase							DMD ↑ 0.15 ± 0.04 0.10 ± 0.02 -1.58(-2.39 – -0.77)
EMG-PS rectus femoris swing phase							DMD ↑ 0.18 ± 0.06 0.12 ± 0.03 -1.26 (-2.04 – -0.49)
EMG-PS medial hamstrings swing phase							DMD ↑ 0.16 ± 0.05 0.11 ± 0.02

							-1.31 (-2.09 – -0.54)
EMG-PS tibialis anterior swing phase							DMD ↑ 0.22 ± 0.05 0.13 ± 0.03
EMG-PS gastrocnemius - soleus swing phase							-2.18 (-3.07 – -1.29) DMD ↑ 0.08 ± 0.01 0.04 ± 0.01
Coactivation rectus femoris – medial hamstrings as % of max amplitude							-4.00 (-5.22 – -2.78) DMD ↑ 0.15 ± 0.06 0.09 ± 0.03
Coactivation gastrocnemius – tibialis anterior as % of max amplitude							-1.26 (-2.04 – -0.49) DMD ↑ 0.14 ± 0.04 0.10 ± 0.03
Coactivation gastrocnemius-soleus stance [?]	DMD late ↑ <i>Insufficient data</i>						-1.13 (-1.89 – -0.37)

Abbreviations in alphabetic order: DMD = Duchenne muscular dystrophy; EMG-PS = electromyographic profile score; GC = gait cycle;

KeR-EGI= Kerpape-Rennes EMG-based Gait Index (Brevet et al. (2013); max = maximal; TD = typical developing;