KU LEUVEN

FACULTEIT BEWEGINGS- EN REVALIDATIEWETENSCHAPPEN

Movement protocol in children with dyskinetic cerebral palsy: a comprehensive approach

door Ellen VAN WONTERGHEM
masterproef aangeboden tot het behalen van de graad van Master of Science in de revalidatiewetenschappen en kinesitherapie

en Sophie GARDYN
masterproef aangeboden tot het behalen van de graad van Master of Science in de revalidatiewetenschappen en kinesitherapie

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prof. dr. E. Monbaliu, promotor
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Opgesteld volgens de richtlijnen van de *Gait & Posture*

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Woord vooraf

In het kader van het behalen van een diploma master in de revalidatiewetenschappen en kinesitherapie aan de Katholieke Universiteit te Leuven werd onderzoek gevoerd binnen het domein van dyskinetische cerebrale parese. Het werk dat voor u ligt is het resultaat van twee jaar onderzoek en werd geschreven door Van Wonterghem Ellen en Gardyn Sophie. Voor we van start gaan wensen we nog enkele cruciale personen uitvoerig te bedanken zonder wie dit werk onmogelijk was.

Bedankt allen.

Dank professor Monbaliu, dank Inti en dank Marco voor de vele antwoorden op ontelbare vragen. Bedankt om ons te begeleiden door dit fascinerende onderwerp en ons te leiden in het pad dat de zoektocht vormde naar de beste versie van onszelf. Dankzij jullie evolueerden wij tot meer kritische individuen in het werkveld, onderzoek en in de maatschappij. Dank voor de leuke samenwerking.

Dank aan alle participanten die deelnamen aan deze studie. Dank aan de ouders voor het vrijmaken van tijd en het maken van de verplaatsing.

Dank aan onze ouders, om ons te vormen tot wie we zijn, bij te staan en graag te zien. Bedankt voor jullie grenzeloos relativeringsvermogen en steun in elk dal. Bedankt voor jullie bewondering op elk hoogtepunt. Bedankt voor de onverbiddelijke warmte en stuwkracht die wij hebben mogen ervaren van kleuter, tot lagere school tot universiteit, en tot waar onze paden ons nog mogen leiden.

Dank allen die de voorbije vijf jaar ons pad doorkruisten. Bedankt vrienden, voor de fijne momenten die we samen mochten beleven. Afstuderen wordt geen einde, onze paden zullen blijven kruisen.

Dank Sophie, voor je mateloze geduld, nuchtere verstand en doorzettingsvermogen. Samen doorzwommen we een soms loodzware en vooral plezante studententijd in West-Vlaamse wateren en Brabantse meren, een masterproef en pandemie. Bedankt voor het delen van dit avontuur, elke lach en traan. Ik had me geen betere thesispartner kunnen wensen. Boven alles, oneindig veel dank voor deze geweldige vriendschap en vele herinneringen.

Dank Ellen, voor je vertrouwen, je motivatie en drijfkracht. We maakten samen een reis doorheen onze studentenjaren met obstakels, maar ook veel positieve, leuke en memorabele momenten. We schreven samen aan een hoofdstuk in onze levensverhaal met onze masterproef als apotheose. Bedankt mij te stimuleren tot het uiterste te gaan en de beste vertolking van mezelf te helpen presenteren.

Hierop volgend wensen we jullie graag veel leesplezier bij het doornemen van dit manuscript.

Eeklo , 21 mei 2021 E.V.W.

Deerlijk, 21 mei 2021 S.G.

Situering

Deze masterproef met als titel 'Movement protocol in children with dyskinetic cerebral palsy: a comprehensive approach' kadert binnen het behalen van het diploma van een masteropleiding in de revalidatiewetenschappen en kinesitherapie aan de Katholieke Universiteit te Leuven. Het kan binnen het grotere geheel gesitueerd worden binnen onderzoek in de onderzoeksgroep Neurorevalidatie (eNRGy) aan de faculteit Bewegings- en Revalidatiewetenschappen.

De masterproef kwam tot stand mits continue supervisie van het onderzoeksteam Neurorevalidatie aan KU Leuven campus Brugge, met aan het hoofd hiervan prof. dr. E. Monbaliu. De focus van dit team ligt op breed onderzoek binnen de populatie van kinderen met dyskinetische cerebrale parese. De aangaande studie uitgevoerd binnen deze masterproef maakt meer specifiek deel uit van het doctoraatsproject van dra. I. Vanmechelen, met de titel 'Instrumented dystonia and choreoathetosis assessment protocol (IDCA) of upper limb movements in cerebral palsy'.

Dyskinetische cerebrale parese (DCP) is de tweede meest voorkomende groep binnen de populatie van kinderen met cerebrale parese. Twee dominante bewegingsstoornissen die we onder het eerstgenoemde kunnen onderscheiden zijn dystonie en choreoathetose. Deze bewegingsstoornissen komen vaak simultaan voor en hebben een grote impact op het dagelijkse functioneren van de kinderen. Ondanks de zware impact van DCP op het dagelijks functioneren, staat het onderzoek naar specifieke behandelingen nog in zijn kinderschoenen. Op dit moment worden de therapeutische behandelingen gebaseerd op klinische ervaring, zonder een toereikende onderbouw van wetenschappelijke literatuur. Medicamenteuze therapie wordt bemoeilijkt gezien sommige medicatie werkzaam voor dystonie, choreoathetose net erger maakt, en omgekeerd. Het kunnen evalueren en discrimineren van beide bewegingsstoornissen is van groot belang. Er werden reeds klinische schalen ontwikkeld om dystonie en choreoathetose te evalueren en te discrimineren, maar deze hebben echter ook limitaties. Ze zijn namelijk deels subjectief, tijdrovend en bovendien wordt een bepaalde ervaring van de beoordelaars verwacht. Drie-dimensionele bewegingsanalyse kan een mogelijkheid bieden om deze bewegingsstoornissen op een objectieve manier te evalueren en te discrimineren. In de literatuur werd eerder reeds gebruik gemaakt van markers en sensoren om bewegingsstrategieën in verschillende pathologieën te analyseren. Vorig jaar werd het Instrumented Dystonia and Choreoathetosis Assessment (IDCA) protocol ontwikkeld, een betrouwbaar en valide protocol dat ons in staat stelt bewegingen in het bovenste lidmaat bij kinderen en jongeren met DCP objectief te evalueren met behulp van marker en sensor data analyses.

Deze studie bouwt verder op dit protocol en heeft als doel gebruikmakend van deze objectieve metingen kenmerkende parameters te vinden om een automatische discriminatie van dystonie en choreoathetose te realiseren.

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Abstract

BACKGROUND: Dystonia and choreoathetosis are two motor disorders in dyskinetic cerebral palsy (DCP). Discrimination and evaluation is difficult, impact on functional activities is large and optimal management is challenging. Towards delineated treatment, a better objective differentiation is needed.

RESEARCH QUESTION: Can 3D upper limb measurements be used to differentiate dystonia and choreoathetosis in an automated manner within the DCP population?

METHODS: In this cross-sectional study, 12 children with DCP and MACS level I-III were evaluated during three functional reaching tasks. The Dyskinesia Impairment Scale (DIS) was used to score dystonia and choreoathetosis in the distal arm. Wrist kinematics and forearm accelerometer and gyroscope data, obtained by optical motion capture and an inertial measurement unit, were implemented as input in the support vector machine (SVM). The predicted classes of the SVM were validated against the DIS classifications of the functional tasks, using accuracy to indicate the model performance. Subsequently, a feature selection procedure was performed to identify the relevant characteristics for each motor disorder. Spearman's correlation and intraclass correlation coefficients (ICC) additionally assessed the validity and reliability of the DIS in this protocol.

RESULTS: The SVM multiclass classifier with 23 features yielded 74.58% accuracy. For the binary classifiers, 22 and 21 features were respectively identified for dystonia and choreoathetosis with 90.34% and 84.66% accuracy. Spearman's correlation and ICCs ranged respectively from 0.648 to 0.830 (p<0.05), and 0.649 to 0.772 (95% CI [0.503-0.838]) for the dystonia subscale, whereas the choreoathetosis subscale results ranged respectively from 0.046 to 0.580 (p=0.030-0.870) and 0.076 to 0.671 (95% CI [-0.069-0.762]).

SIGNIFICANTS: This study was the first to validate automated dystonia and choreoathetosis discrimination during functional tasks. The promising outcomes can assist treatment management, thereby improving quality of life in patients with DCP. Future research should enhance the instrumented dystonia assessment tool and clinical evaluation of choreoathetosis.

List of abbreviations

CP	Cerebral Palsy
SCPE	Surveillance of Cerebral Palsy in Europe
DCP	Dyskinetic Cerebral Palsy
DYS	Dystonia
CA	Choreoathetosis
MACS	Manual Ability Classification System
GMFCS	Gross Motor Function Classification System
MRI	Magnetic Resonance Imaging
BADS	Barry Albright Dystonia Scale
DIS	Dyskinesia Impairment Scale
ULEMA	Upper Limb Evaluation in Motion Analysis
MOCAP	Optical Motion Capture
IMU	Inertial Measurement Unit
IDCA	Instrumented Dystonia and Choreoathetosis Assessment
RF	Reach Forward
RGV	Reach and Grasp Vertically
RS	Reach Sideways
PTA	Point of Task Achievement
ROM	Range of Motion
A / ACC	Accelerometer data
G / GYR	Gyroscope data
RMS	Root Mean Square
SD / STD	Standard Deviation
SVM	Support Vector Machine
RBF	Radial Basis Function
OvO	One-versus-one encoding multiclass classifier system
ICC	Intraclass correlation coefficient
CI	Confidence interval
Med	Median
VAR	Variance
MAX	Maximum
J	Jerk

SE	Sample entropy
COR	Correlations between axes
MAXVEL	Maximal linear velocity (ACC)
SM	Smoothness
SpE	Spectral entropy
FbandP	Power in frequency band x
TrDev	Trajectory deviation
Vmax	Maximal linear velocity (MOCAP)
WrFlex	Wrist flexion
WrDev	Wrist deviation
Ct.	Category

1. Introduction

“Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture, causing activity limitations, that are attributed to non-progressive disturbances occurring in the developmental fetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, and behavior, by epilepsy, and by secondary musculoskeletal problems.”[1] CP is one of the most common causes of physical disability in children.[2] According to the Surveillance of Cerebral Palsy in Europe (SCPE), the condition occurs in 1.5-3 per 1000 live births.[3]

Since CP causes variable symptoms, the condition is classified according to the types of motor disturbances and the specific brain lesions. The SCPE classified CP into three groups: spastic, dyskinetic and ataxic cerebral palsy.[4] The dyskinetic CP population is the second most prevalent group and has an incidence varying between 6.5% and 14.4%.[2, 5, 6] This variation is attributed by a lack of standardization regarding the definition and classification based on the predominant type.[3, 4]

Dyskinetic cerebral palsy (DCP) is characterized by involuntary excessive and repeated movements. The two dominant motor disorders are dystonia and choreoathetosis. Dystonia (DYS) is presented by twisting and repetitive movements, abnormal postures due to sustained muscle contractions, and hypertonia. Choreoathetosis (CA) is characterized by hyperkinesia (chorea i.e. rapid involuntary, jerky, often fragmented movements) and hypotonia (athetosis i.e. slower, constantly changing, writhing or contorting movements).[4, 7] Dystonia and choreoathetosis mostly occur concurrently, but must be seen as separate entities.[7]

Clinically, patients with DCP are most likely to have a high level of functional disability and more accompanying impairments, compared to the other subtypes of CP.[8-10] Around 60% of the children with DCP are classified in the severe levels of functional disability in gross motor function, underscoring the importance of manual ability in maintaining their autonomy and participation. However, only 36.4% of the children have an adequate manual ability (Manual Ability Classification System (MACS) level I-III).[11] Additionally, research revealed an association between higher levels of dystonia and a more severe motor function, unlike choreoathetosis in which no associations were found. This suggests that dystonia has a larger impact on functional ability than choreoathetosis.[12]

Apart from the clinical classification, multiple classification systems using Magnetic Resonance Imaging (MRI) have been developed, linking brain lesions to time of birth, CP subtype and functional ability, to elucidate the pathological aspects of CP.[13-16] Around 70% of patients with DCP shows lesions in the basal ganglia and thalamus. However, other brain lesions and even normal-appearing MRI findings can occur.[12, 15]

Several studies unfortunately demonstrated ambiguity discriminating between the subtypes of CP as well as DYS and CA based on specific lesions with the current MRI technology.[17, 18]

Notwithstanding the necessity, the current treatment management for children with DCP is expensive and only symptomatic. Management is multidisciplinary and consists of pharmacology, surgical and rehabilitation management. The scarce amount of research on the effect of the different interventions in the DCP population shows low efficacy.[19] To decrease dystonia oral drugs, intrathecal baclofen, botulinum toxin injections and surgical options such as deep brain stimulation can be used, but the responsiveness is variable.[17, 19-22] The effects of these treatment modalities have not yet been investigated in choreoathetosis. The importance of differentiating DYS and CA is crucial to ensure a targeted intervention, as misinterpretation in diagnosis can contribute to the administration of inappropriate medication. Anticholinergics e.g. are used to decrease dystonia but can cause worsening of choreoathetosis.[12, 17, 23] Orthopedic surgery is the last medical treatment option after considering all other alternatives.[24] Rehabilitation interventions for DYS and CA are mostly based on clinical expertise with low evidence and are guided by a multidisciplinary team of physiotherapists, occupational therapists and speech therapists.[17]

To date, the evaluation of the severity of DYS and CA in patients with DCP involves the use of clinical qualitative measurement scales judged by video observation based on consensus definitions. Stewart et al. showed that most scales demonstrate a moderate validity and reliability, and also a lack of connection with the current dystonia definition.[21, 22, 25-28]

The Barry Albright Dystonia Scale (BADS) and the Dyskinesia Impairment Scale (DIS) are most commonly used in patients with DCP. The latter is currently stated as the most sensitive, valid and reliable scale. Its additional value lays in its detailed full-body consideration and the differentiation action-rest, proximal-distal limb and duration-amplitude. Moreover, the DIS is characterized by the inclusion of both DYS and CA evaluation. Although this scale is currently seen as the gold standard in evaluating DYS and CA in patients with DCP, substantial time and experience with the current DYS and CA definitions is needed from the rater.[27-29]

Objective measurement techniques can overcome the limitations of these partly subjective clinical scales in the rating of the DYS and CA severity.[30] Last decades, research focused on the ability to objectify movement characteristics using advanced technologies such as optical motion capture and inertial measurement units. Both techniques have already been used to evaluate movement characteristics in other populations with neurological disorders.[31-40]

Optical motion capture (MOCAP) is the registration of movements in 3D using infrared radiation cameras to detect light-reflecting body-placed markers. Biomechanical characteristics such as joint angles and movement velocities can be calculated from the obtained 3D trajectories.[31]
This approach showed good reliability in the objective measurement of motor function in typical developing children; and also in populations with hypokinetic rigid syndromes, dyskinesia, obstetrical brachial plexus palsy, and hemiplegic spastic CP in the upper limb motion analysis.[31-35] The Upper Limb Evaluation in Motion Analysis (ULEMA) protocol was recently developed for the spastic CP population to standardize the upper limb marker placement and tasks.[41] Although this marker approach has been frequently used for clinical and research purposes, MOCAP has some limitations. The most important limitation is the location-restriction, due to the need for a specifically equipped laboratory. Secondly, this method requires time and experience because of the need for a correct anatomical marker placement, the high number of markers, and their possible displacements due to inadequate fixation.[42]

Apart from MOCAP, biomechanical parameters of 3D movements can be obtained by Inertial Measurement Units (IMUs); which contain three types of sensors: an accelerometer, a gyroscope and a magnetometer.[43] IMUs have frequently been used in research for biomechanical measurement in Parkinson and stroke.[36-40] Benefits promoting the use of the IMUs include the affordability, usability in time and space, and potentially a better data quality for more derived parameters.[44, 45] Arm movements in children with DCP demonstrate a high variability, which will profit from an accurate measurement of more detailed movement characteristics such as acceleration and its deviated jerk values.[46]

Despite the potential of MOCAP and IMU systems in the DCP population, research is currently limited. Gordon et al. explored tonus deficits and reaching performance in both the spastic and dystonic CP population using the MOCAP optometric system. Severe dystonia could be related to a large degree of overflow performing a rest-tap task. Reaching in severe dystonia was characterized by more curved arm trajectories.[47] Sanger et al. investigated the upper limb kinematics using magnetic position sensors. Results indicated an existing random variability and a lack of straight-line pattern in arm trajectories in dystonia. This study demonstrated the ability to distinguish typical developing children from a DCP population using a signal-to-noise ratio, jerk and the index of curvature. In addition, these measures have been suggested to be useful as a quantitative parameter of severity.[46]
At present, limited studies focused on the usage of the IMUs in the evaluation of DYS and CA in the DCP population. Cuyvers et al. studied children with DCP classified in Gross Motor Function Classification System (GMFCS) and MACS level IV and V, who were evaluated for involuntary movements during a head/foot staired wheelchair mobility task.[48] No studies have explored the usage of IMUs in children with DCP with a more adequate manual ability (MACS level I-III).

To date, a more sensitive diagnostic system with objective measurements of the different motor disorders during voluntary movements is lacking in patients with DCP, and hampers targeted treatment management. Objective measurement methods in patients with DCP will enhance insights in movement patterns of this complex disorder. On the long term, this approach could assist clinicians in ameliorating the patient's quality of life by specifying targeted therapy according to the severity of each motor disorder.
Previous research has developed an Instrumented Dystonia and Choreoathetosis Assessment (IDCA) protocol using MOCAP and IMUs to obtain objective characteristics of the upper limb functional movements. This protocol showed good feasibility and validity in distinguishing typically developing children and children with DCP.[49]

Building on this above-mentioned protocol, the ultimate aim of this study is to discriminate DYS and CA in patients with DCP based on instrumented upper limb measurements. The research question is: Can 3D upper limb measurements be used to differentiate dystonia and choreoathetosis in an automated manner within the DCP population? The hypothesis is that jerk and higher frequency-related parameters will be more related to hyperkinetic movements as seen in choreoathetosis, whereas deviating position measures such as joint angles will be more related to abnormal postures as seen in dystonia. As a sub-aim, this study critically explored the reliability and validity of the scoring of the DIS implemented in the functional tasks. The hypothesis is that the DIS is a reliable and valid clinical instrument to evaluate DYS and CA in the IDCA protocol.

2. Materials and Methods

2.1 Participants

This study included 12 Flemish children with DCP, recruited via the University Hospitals Campus Gasthuisberg and Pellenberg, and via KOMPAS (a Flemish organization of special education schools). Inclusion criteria were: aged between 6 and 25 years old, a MACS level I-III and a sufficient cooperation to execute the requested reaching and grasping tasks. Exclusion occurred in case of: other neurological disorders, botulinum toxin type A injections within six months prior to the measurements and neurological or orthopedic surgery within one year. Ethical approval was obtained from the Ethical Committee of the KU Leuven (*Appendix D*). All participants and/or their parents provided written informed consent.

2.2 Study design

This study targeted an objective discrimination of DYS and CA within the DCP population, using an observational cross-sectional study design. Therefore, a reliability and two types of validity studies were created. A discriminative validity study evaluated the predictive outcome of a machine learning model compared to the DIS classification scores of the functional tasks. An additional study assessed the concurrent validity by comparing the DIS score percentages of DYS and CA for the tasks of the DIS with DIS score percentages for the functional upper limb tasks. Moreover, the interrater reliability of the DIS scoring in the functional tasks was investigated. Further elaboration of these studies will be discussed in the next sections.

2.3 Data collection

2.3.1 Material

Participants were asked to perform three functional tasks: reaching forward (RF), reach and grasp vertically (RGV) and reaching sideways (RS). A chair and a custom-made reaching system were developed to enable adjustments according to the arm length and shoulder height of the participant.[41]

Joint kinematics for the upper limb movements were obtained using MOCAP and IMUs. This data collection took place at the fully equipped clinical motion analysis laboratory situated in KU Leuven Campus Bruges and the University Hospital in Pellenberg.

The MOCAP system contained 31 markers registered by 12 infrared radiation cameras of the VICON motion capture system (Oxford Metrics, UK), sampled at 100Hz. Seventeen physical segmental markers, clustered on tripods and cuffs, were placed on the shoulder, arm and hand according to the ULEMA protocol.[41] Fourteen additional anatomical landmarks were captured in the static sitting position using a marked pointer. The palpated landmarks allowed for calculation of coordinates for four joint centers: the glenohumeral and scapulothoracic joint, the elbow and wrist joint.[50] Additionally, five IMU devices including accelerometers and gyroscopes (Xsens, The Netherlands) were placed on the hand, forearm, upper arm, scapula and sternum, based on the IDCA protocol.[49] The IMUs were time-synchronized with the VICON MOCAP system. The placement of the markers and the IMUs is shown in *figure 1*.

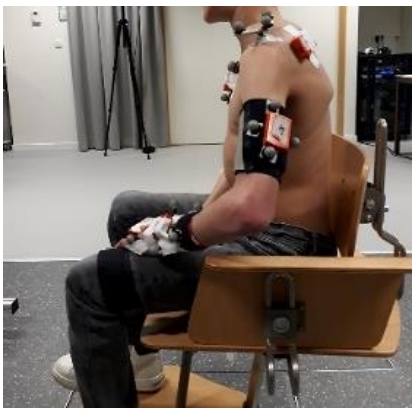


***Figure 1**: Placement of markers and IMU devices*

The DIS was included as the gold standard to evaluate and discriminate DYS and CA clinically. More details of the full scoring system are illustrated in *figure 2*.[27]

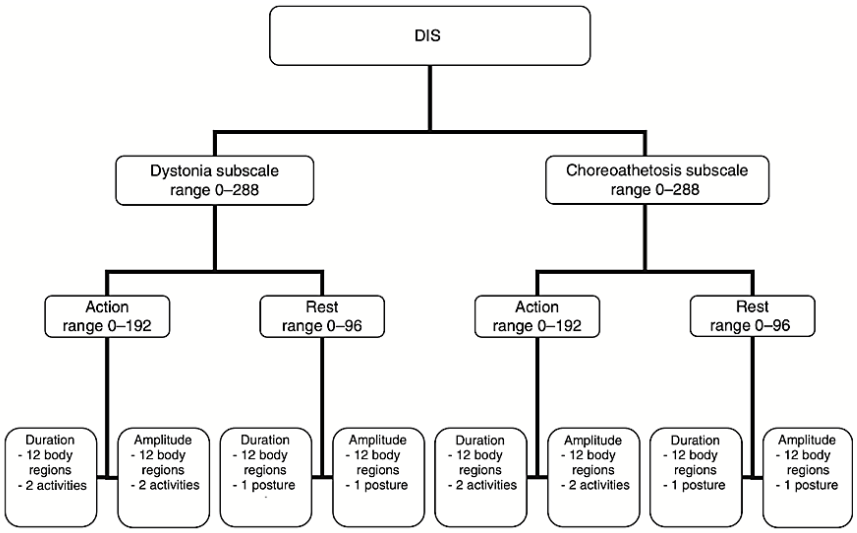


***Figure 2**: The Dyskinesia Impairment Scale*

2.3.2 Protocol and test set-up

Each test was structured as followed:

Static and dynamic calibrations of the MOCAP and IMU systems were executed in the reference position, a neutral sitting position with the hands placed on the ipsilateral knee.[41] The palpated anatomical landmarks were digitized using a marked pointer.[41]

The participants subsequently performed the three selected functional reaching tasks with the most affected arm, at a self-selected speed. In case both arms were affected, they were included as different subjects. Each task was performed three times, with ten repetitions each. One repetition is defined from the reference position to the target position (point of task achievement, PTA). The tasks are illustrated in *figure 3.* These tasks were selected because of their challenging and differentiating attributes, taking into account the functional possibilities of children with DCP.[49]

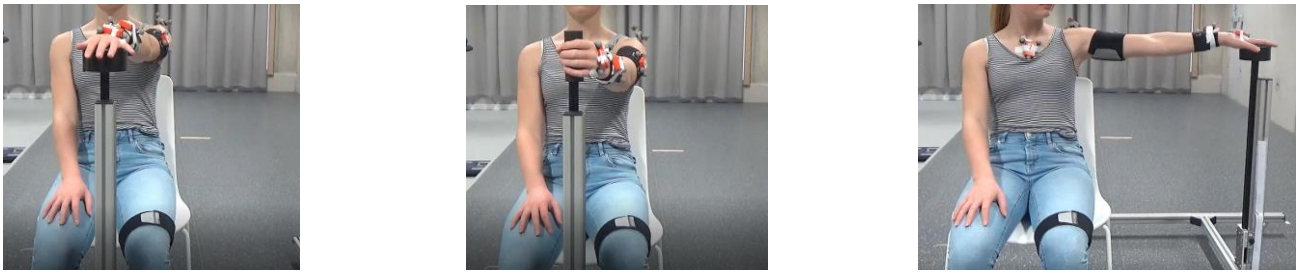


Figure 3*: Test protocol (RF-RGV-RS)*

A subset of the DIS tasks (i.e. proximal – distal arm) as well as the functional reaching tasks were captured through video recordings. The subset of the DIS tasks contained arm abduction, reach and grasp a pen in supine position, reach and grasp a cup, and reach and grasp a pen in sitting position. Each DIS task comprised ten repetitions, which have been scored all at once. In contrast to the DIS tasks, all functional reaching tasks were scored per repetition separately. Due to the short duration of each of these repetitions (usually ranging between 1 and 4 seconds), duration was scored on a 3-point instead of a 5-point scale; with 0 = no DYS/no CA, 1 = less than 50% of the time DYS/CA and 2 = more than 50% of the time DYS/CA. The amplitude of the dystonic and choreoathetotic movements were scored on a 5-point scale as in the DIS; with 0 = no DYS/no CA, 1 = DYS/CA in a small ROM (<10%), 2 = DYS/CA in a moderate ROM (≥10 - <50%), 3 = DYS/CA in a submaximal ROM (≥50 - <90%), 4 = DYS/CA in a maximal ROM (≥90%). Finger movements were excluded, as it was not possible to place an IMU device on the fingers and since reliable, comfortable and natural placement of the markers on the interphalangeal heads is challenging. Finger measures were therefore also excluded in the distal arm DIS evaluation.

The test procedure was administered by two assessors educated in rehabilitation and movement science and with adequate knowledge about the administration of the DIS and the IDCA protocol. The assessors were not blinded and no randomization of the selected subjects occurred.

2.4 Classification distribution

Consensus scores were obtained after a revision of the DIS scores by both assessors. The DIS outcomes for the functional tasks, serving as reference in the automatic classification, were categorized into four score classes for DYS and CA. The classifications served as input in the machine learning model. The definitions of the classifications are presented in *table 1.*

***Table 1**: Four-class DIS classification system*

CLASS	DEFINITION
0	No DYS/No CA
1	DYS/No CA
2	No DYS/CA
3	DYS & CA

After scoring and classifying the functional reaching tasks, the distribution of the DIS classification data was analyzed, as illustrated in *figure 4*. An uneven dispersion was obtained, with a predominant presence of observations in class 1 (DYS/No CA). Due to this low variability, especially in the proximal arm classification, the DIS and machine learning data analysis for the proximal arm were excluded from this study. Hence, only parameters related to the distal arm were included. After in depth analysis of the data, missing data for the hand IMU and the need to limit the initial feature input led to the decision to only include the sensor data of the wrist. This study thus included the forearm features for the IMU and the wrist and hand kinematics for the MOCAP system.

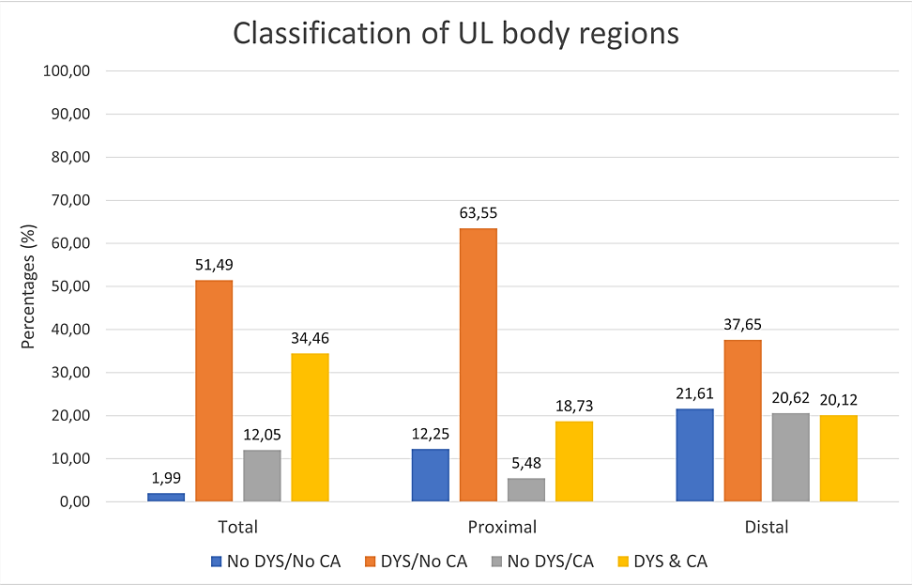


***Figure 4:** Overview of the distribution of the DIS classification scores in the total arm, the proximal arm and the distal arm. Represented as a percentage of the total observations in class 0-3.*

2.5 Data analysis

2.5.1 Preprocessing data

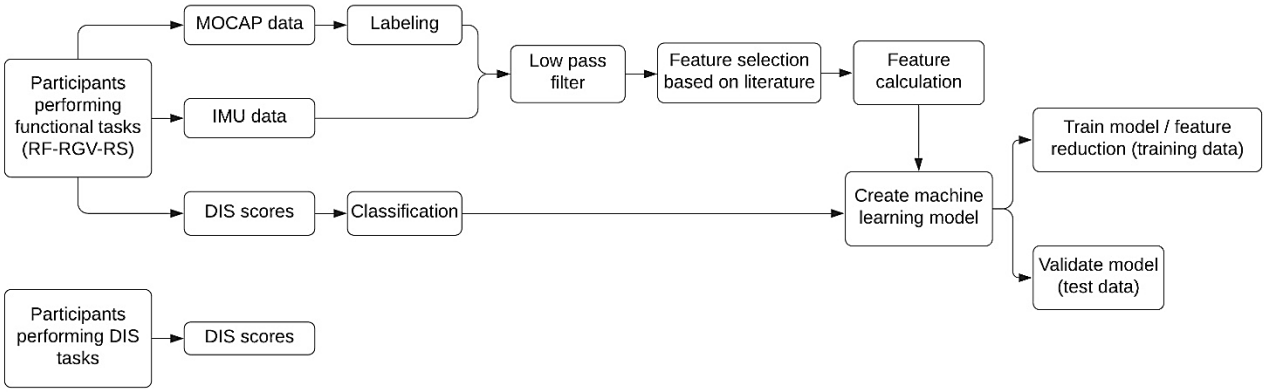


***Figure 5**: Flowchart of the data analysis*

The data analysis process is depicted in *figure 5.* The first step in the data analysis was to pre-process the data in VICON and MATLAB (MATLAB 8.6.0 2019b, The MathWorks Inc., Natwick, MA, USA). An example of the raw MOCAP and IMU data is illustrated in *figure 6*. Due to weak signal transmission, loss of data and errors in the data collection occurred. For the MOCAP, incorrect joint angles due to marker occlusions were omitted from the dataset. All data from the IMUs was checked and corrected where necessary (e.g. corrupt sensor signals). Gaps in the MOCAP kinematic data were filled using a spline-based fill and the data was filtered with a spline-based Woltring filter. IMU signals were filtered with a low-pass Butterworth filter (4^{th} order) with a cut-off frequency of 5Hz.

From the time-dependent signals in *figure 6*, time-independent variables were calculated and included in the feature selection (*section 2.5.2*) to fed in a machine learning classifier.

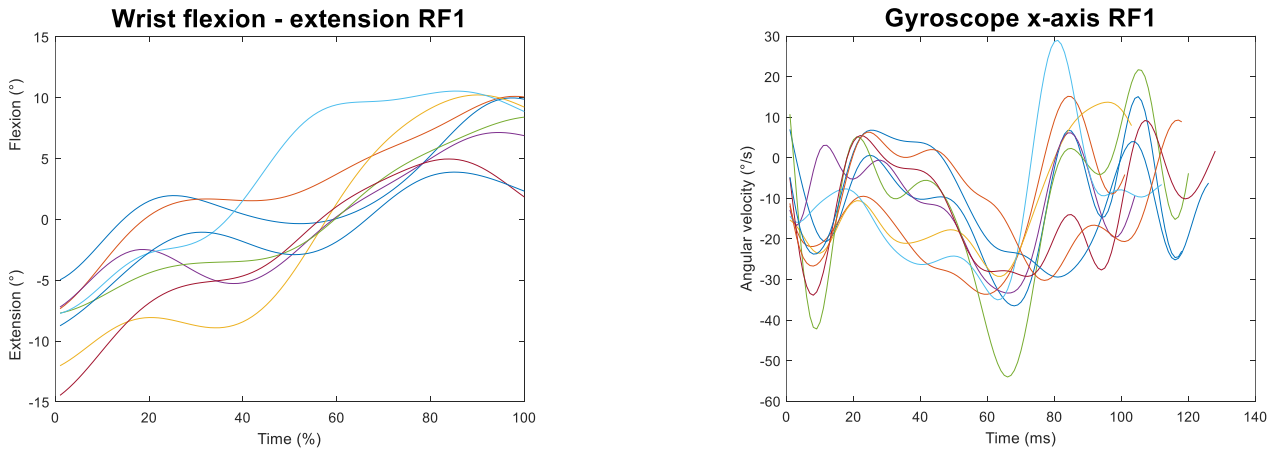


***Figure 6:** Illustration of the raw data: a) the MOCAP data, wrist flexion and extension joint angles (°) during RF1 task; b) the IMU data of the forearm, gyroscope values (°/s) of the x-axis during RF1 task. One waveform represents one repetition.*

2.5.2 Feature selection based on literature

In the DCP population, excessive and jerky movements of multiple joints are observed in reaching tasks. These can be seen distally in a poorly controlled hand posture, orientation and end-point force, and contralateral mirroring.[46] Though, qualitative findings revealed differences in distribution for DYS and CA across the different body regions.[12] Moreover, dystonia was found to be mainly characterized by more defined translational movements (i.e. repetitive abnormal postures, twisting movements), whereas choreoathetosis showed more noisy, complex and chaotic rotational movements (i.e. jerky, involuntary movements).[48](personal communication)

Hypertonic characteristics can be identified using features such as joint angles, average velocity, spectral arc length, and jerk, when compared to a healthy population.[51-54] For hyperkinesia, mean and standard deviation of the acceleration signal and sample entropy were found to be discriminative compared to a healthy population.[55, 56] As hypertonia is a prominent characteristic for dystonia, and choreoathetosis is manifested with hyperkinesia, the previous features were of interest in this study.

For the comparison of DYS and CA in a state of relative rest, accelerometer features were found to predominantly delineate dystonia, whereas gyroscope features predominantly delineated choreoathetotic movements. Spectral entropy of the angular velocity signal characterized the continuously changing choreoathetotic movements. Ambiguity existed about the presence of significant higher power in higher frequency bands in choreoathetosis.[48](personal communication) Due to the inconsistent conclusions of previous studies and the importance of not overlooking relevant parameters, both time and frequency domain features were included in this study. Different results were hypothesized since the outcomes of this prior research could not be compared to this study due to the differences in the target group and test protocol.

Results of previous research together with the consensus definitions contributed to an overview of parameters for discriminating DYS and CA in the IDCA protocol. The selected parameters were split into MOCAP and IMU features, and further into time and frequency domain features for the IMU data. A hypothesis on how these features relate to DYS and CA is also included in *table 2* and *3* below.

All selected upper limb kinematic features were calculated from the 3D ULEMA protocol in MATLAB.[57] A total of 144 (6 MOCAP + 138 IMU) time and frequency domain features were extracted for further data analysis.[57]

2.5.2.1 Optical motion capture

An overview of the features obtained from the MOCAP system is presented in *table 2*. Minimum and maximum angles were calculated from the joint angles over time, using the ULEMA protocol in MATLAB.[57] Maximal linear velocity was obtained by calculating the derivative of the displacement over time of the marker placed on metacarpal III. Trajectory deviation represents the sum of the deviations of that same marker from the linear constant taken between two points in motion.

Table 2*: Time domain features obtained from the MOCAP system*

TIME DOMAIN FEATURES	No. OF FEATURES	RELATED SYMPTOM
Joint angles Min, max	4	DYS
Linear velocity Max	1	DYS
Trajectory deviation	1	CA

2.5.2.2 Inertial measurement units

Features included from the IMU are listed in *table 3*. First, time domain features are clarified. Linear velocity was obtained by integrating the acceleration values over a specific time interval. Linear jerk is defined as the derivative of linear acceleration and angular jerk as the second derivative of angular velocity. Jerk is also an indication of the smoothness of movements in the time domain. The sample entropy assesses the complexity and regularity within the time domain data, as it measures the predictability of further data points based on a given data point. Lastly, a pairwise correlations between the x/y/z axes in both accelerometer and gyroscope were calculated with Pearson correlation coefficients. Duration describes the time needed to perform one repetition, from the reference point till the target position.

Subsequently, frequency parameters are of interest in interpreting subtle differences in movement patterns. These parameters have been extrapolated after Fourier transformation of acceleration and angular velocity signals into the frequency domain. Smoothness, in the frequency domain, can depict the amount of change in this signal frequency data. The spectral arc length is a standard for the evaluation of smoothness and calculates the length of the curve in the Fourier power spectrum of a speed profile. It is the most valid, sensitive, consistent and robust smoothness measure.[58] Another measure to calculate the complexity in frequencies is the spectral entropy, estimating the uniformity of the signal energy distribution. Power represents the strength/energy of a signal in a certain frequency band.

***Table 3**: Time and frequency domain features obtained from the IMU*

	No. OF FEATURES	RELATED SYMPTOM
TIME DOMAIN FEATURES		
Angular velocity x/y/z/norm (GYR)	16	DYS
Max, RMS, SD, variance		
Linear velocity x/y/z (ACC)	3	DYS
Max		
Acceleration x/y/z/norm (ACC)	16	DYS
Max, RMS, SD, variance		
Angular jerk x/y/z/norm (GYR)	16	CA
Max, RMS, SD, variance		
Linear jerk x/y/z/norm (ACC)	16	CA
Max, RMS, SD, variance		
Sample entropy x/y/z/norm (ACC & GYR)	8	CA
Angular velocity – acceleration		
Correlation between axes (ACC & GYR)	6	DYS-CA
Duration	1	DYS
FREQUENCY DOMAIN FEATURES		
Smoothness: Spectral Arc Length (ACC & GYR)	8	CA
Angular velocity – acceleration		
Spectral entropy (ACC & GYR)	8	CA
Angular velocity – acceleration		
Power in frequency bands (ACC & GYR)	40	DYS / DYS / DYS-CA /
0-0.5 Hz / 0.5-1 Hz / 1-2 Hz / 2-3 Hz / 3-4 Hz		DYS-CA / CA

ACC = calculated from the accelerometer data; GYR = calculated from the gyroscope data

2.5.3 Machine learning

Because of the need for an adequate and easy-to-use classification of the motor disorders, the analysis of data and statistics was processed using machine learning techniques in MATLAB. Machine learning is a part of artificial intelligence and is based on the idea that algorithm models can learn from training data without being explicitly programmed. For this study, the support vector machine (SVM) was the best fitted machine learning classifier model, induced by the capacity to learn from a smaller dataset (N ≈ 1000) and high dimensional data (No. of features = 144).

The full dataset for this study included the MOCAP and IMU feature input for the 948 observations available. First of all, the dataset was randomized and split into a 75% training and 25% test dataset. The training data was labeled; i.e. each observation was attributed a DIS classification score. The small training dataset of this study led to the necessity to utilize cross validation, using the 75% of the data for a 10-fold training of the machine learning model. Ten-fold cross validation implicates multiple training of the model with randomly selecting 90% of the training data and validating it with the remaining 10% of the data (i.e. tuning hyperparameters, see *section 2.5.3.1*). Ultimately, the remaining 25% unlabeled data (i.e. no DIS classifications) was retained as test data to validate the SVM model. The power of this model depends on the number of observations related to the number of features. As the 948 observations included in this study were more than double the number of features, the statistical power of this study is considered sufficient.

2.5.3.1 Support vector machine classifier model

The SVM is a supervised learning algorithm which targets to find the best fitted mathematical function to classify DYS and CA in the labeled dataset. More specifically, the algorithm looks for similarities between the classes in the given training data and creates support vectors based on these similarities. Subsequently, the best hyperplane function is calculated based on these support vectors. As high-dimensional, non-linear data was included, kernel functions were required. A kernel function performs a fictional transformation of the dataset into a higher dimension, offering the opportunity to create the hyperplane. In this study, the Radial Basis Function (RBF) kernel function was used, which is recommended due to its ability to deal with irregular datasets and to approximate non-linear functions, its good generalization and fast learning capacity.[59] The position of the hyperplane is based on the trade-off between maximizing the margin between classes/support vectors and minimizing the number of misclassification in the training data. The trade-off of this boundary can be controlled by tuning hyperparameters in training the model, i.e. the c-regularization and gamma (γ) parameter. A large c results in a lower number of misclassifications by minimizing the margin of the hyperplane. A large γ only includes data points close to the hyperplane. The full process to obtain the most appropriate value of hyperparameters was realized by 10-fold cross validation, which was specified in the section above.

In the first stage, the full dataset was implemented to enable automatic discrimination of the four DIS classes with each other (*table 1*). Since more than two classes were compared, a multiclass classifier system using one-versus-one (OvO) encoding was implemented. Hence, six models were created to binary compare the classes with each other (i.e. 0-1 / 1-2 / 2-3 / 0-2 / 0-3 / 1-3). The cross-validation process was run ten times for all six models. The multiclass classifier and the decision boundaries are visualized in two dimensions in *figure 7*. Using this implementation, manual feature reduction was processed, which will be discussed in the following section.

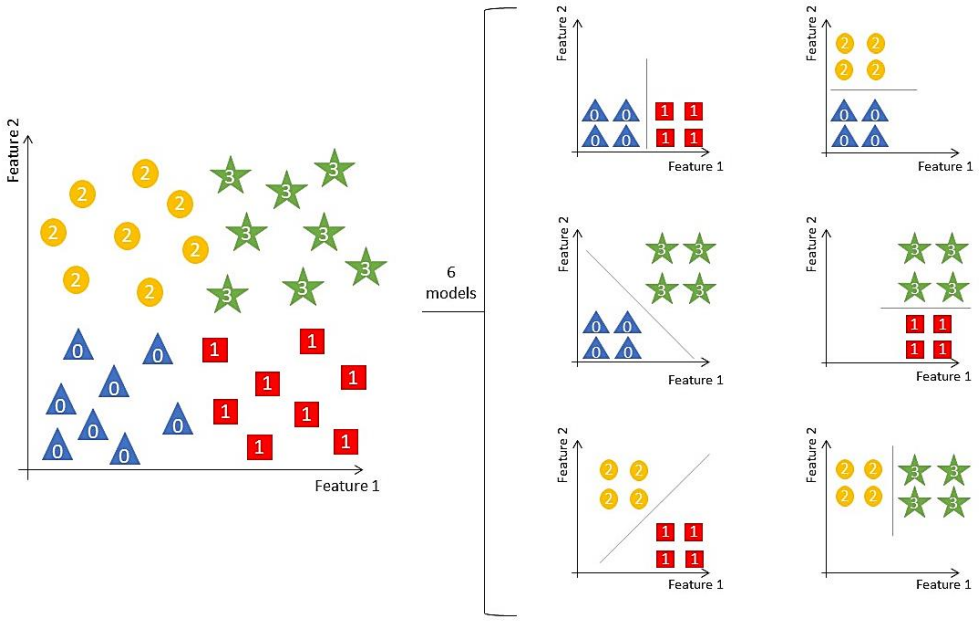


***Figure 7**: Visualization in 2D: one-versus-one multiclass classification (six submodels i.e. 0-1 / 0-2 / 0-3 / 1-3 / 1-2 / 2-3). The full line represents the linear decision boundary for each model.*

In the second stage, the full dataset was split in a dystonia and choreoathetosis dataset i.e. DYS/CA present or not present. This reclassification is visualized in *table 4.* Since only two classes were compared in this stage, a binary classification in the SVM model was implemented with manual feature reduction. Targeted conclusions could be made by objectifying the characteristic features of both motor disorders using this reduction process.

***Table 4:** Reclassification – binary classifier dystonia and choreoathetosis dataset*

CLASSES	DEFINITION	RECLASSIFICATION DYS	RECLASSIFICATION CA
0	No DYS/No CA	**0 (No DYS)**	**0 (No CA)**
1	DYS/No CA	**1 (DYS)**	**0 (No CA)**
2	No DYS/CA	**0 (No DYS)**	**1 (CA)**
3	DYS & CA	**1 (DYS)**	**1 (CA)**

2.5.3.2 Feature reduction

While training the SVM classifier model, feature reduction was implemented on the previously described datasets to optimize the input of the model.

A set of the most relevant features was obtained by repetitive analyses of the accuracy (also F1-measure, precision and recall) of different models, using for instance manual backward and forward sequential feature selection. Backward feature selection starts with the full dataset and leaves features out one-by-one, carefully controlling the accuracy of the model. Forward feature selection starts with the inclusion of one feature, and consistently adds more features to the dataset. This optimization procedure of the input features is a trial-and-error process. Since outcomes of the SVM vary depending on the randomly chosen observations in training, a 10-fold run was opted of which the median outcome was reported. Hereby, controlled decision making in this manual feature reduction process was permitted.

2.6 Statistical analysis

At last, one reliability study and two validation studies were implemented. The initial DIS scores of rater 1 and rater 2, prior to in depth discussion, were analyzed in the interrater reliability study. Concurrent and discriminative validity analyses were completed using the consensus scores of the most experienced rater after in depth discussion.

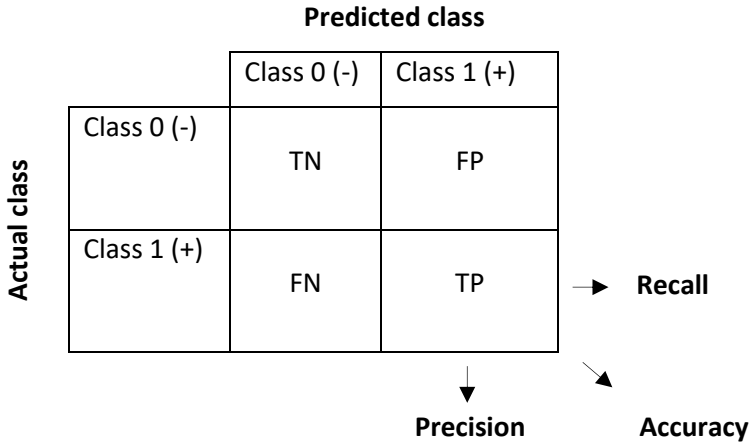
In a first part, the concurrent validity study compared the DIS severity percentages in the DIS tasks with the DIS severity percentages in the functional reaching tasks used in this study protocol. Validation of the DIS in these functional reaching tasks will be important serving as a reference for training and validation in the SVM study. A total of 22 variables was compared (mean and median of DYS and CA: four variables for the total of the three functional tasks – four variables for RF – four variables for RGV – four variables for RS / DYS and CA: two variables for the total and two variables for each task of the DIS). Due to the categorical characteristic of the DIS, a small sample and the absence of a normal distribution in the functional task scores, Spearman's rho correlation coefficients (r_s) were used. Correlation coefficients above 0.75 were considered as excellent; between 0.50 and 0.75 as moderate to good; between 0.25 and 0.50 as fair; and between 0 and 0.25 as no correlation.[60]

In addition, intraclass correlation coefficients (ICC) and the 95% confidence intervals (CI) were calculated to check for agreement in inter-rater scoring (k = 2). The calculations were based on a severity score rating, absolute agreement, 2-way mixed effects model. ICC values above 0.90 were considered as excellent; between 0.75 and 0.90 as good; between 0.60 and 0.75 as moderate and less than 0.60 as poor.[60]

The previous statistical analyses were conducted with SPSS statistical package version 26 (SPSS Inc, Chicago, IL). The level of significance was set at $p<0.05$.

In a second part, a discriminative validity study compared the predictive outcome of the SVM classifier model in classifying DYS and CA during the functional reaching tasks with the actual pre-assigned classifications by the rater. The classifier performance was evaluated by calculating the accuracy, F1-measure, precision and recall of multiple models. Calculation is based on the interpretation of confusion matrices, which represent the actual and predicted classifications. The calculation is illustrated below.

Table 5: *Illustration of the confusion matrix interpretation and the calculation of the performance measures*



$$\textbf{Recall} = \frac{TP}{TP+FN}$$

$$\textbf{Precision} = \frac{TP}{TP+FP}$$

$$\textbf{F1 - measure} = \frac{2*Precision*Recall}{Precision+Recal}$$

$$\textbf{Accuracy} = \frac{TP+TN}{Total}$$

TN = true negative; FN = false negative; FP = false positive; TP = true positive

Accuracy represents the proportion of correctly predicted observations relative to the total observations. In case class distribution is imbalanced, it is interesting to interpret the precision outcome, displaying the percentage of correctly predicted positive observations related to the total predicted positive observations. Recall is a measure of sensitivity which is calculated as the percentage of correctly predicted positive observations related to all observations in the specific class. The F1-measure represents the harmonic mean of the precision and recall features. Finally, the classifier misclassification rates (or prediction errors) were calculated. The misclassification rate enumerates the variance and bias created by the machine learning model (i.e. trade-off error terms *section 2.5.3.1*).

3. Results

3.1 Participants

Twelve children (seven males and five females, mean age 17 years; range 8y 6m to 25y 3m) with DCP were included in this study, in which both arms were evaluated in five participants. One subject was eventually excluded because of missing data, forming a total of 16 subjects. Two subjects manifested with a manual ability classified as level I, eight were situated in level II and six subjects were classified in level III. More details about the participants are presented in *Appendix A, table 1*.

3.2 Dyskinesia Impairment Scale in functional reaching tasks

3.2.1 Concurrent validity

The results of the concurrent correlation analyses for the DIS dystonia and DIS choreoathetosis subscales are presented in *table 6* and *7* respectively. The variables are explained in the caption of these tables.

Significant correlations were found for the complete **dystonia subscale** of the distal arm. The correlations ranged from 0.648 to 0.830 (p=0.000-0.011), showing moderate to excellent relationships. Correlations with the original DIS were highest in the RF tasks, with overall excellent and highly significant results for AD_grasp pen (mean r_s=0.806, p=0.000; med r_s=0.815, p=0.000), AD_total (mean r_s=0.790, p=0.000; med r_s=0.814, p=0.000) and AD_grasp cup (mean r_s= 0.763, p=0.001; med r_s=0.809, p=0.000). RS tasks were more related with AD_grasp cup (mean r_s=0.771, p=0.001; med r_s=0.754, p=0.001).

For the **choreoathetosis subscale**, correlation coefficients ranged from 0.046 to 0.580 (p=0.030-0.870), showing absent to moderate associations. Merely six significant correlations were found for the choreoathetosis subscale, mostly situated in the RGV tasks. RGV_mean showed moderate associations with AD_total and AD_grasp cup (r_s=0.546, p=0.044; r_s=0.561, p=0.037). RGV_med was moderately correlated with all DIS tasks (AD_total r_s=0.567, p=0.034; AD_grasp cup r_s=0.580, p=0.030; AD_grasp pen r_s=0.541, p=0.046). No significant correlations were found for the RF and RS tasks.

***Table 6**: Correlation analyses between DIS score percentages of the DIS tasks and the functional reaching tasks – Dystonia subscale*

FUNCT / DIS	Total_mean		Total_med		RF_mean		RF_med		RGV_mean		RGV_med		RS_mean		RS_med	
	r_s	*p* value	r_s	*p* value	r_s	*p* value	r_s	*p* value	r_s	*p* value	r_s	*p* value	r_s	*p* value	r_s	*p* value
AD_total	0.830	0.000 ***	0.747	0.001 **	0.790	0.000 ***	0.814	0.000 ***	0.689	0.006 **	0.712	0.004 **	0.722	0.002 **	0.684	0.005 **
AD_ grasp cup	0.813	0.000 ***	0.728	0.002 **	0.763	0.001 **	0.809	0.000 ***	0.663	0.010 *	0.679	0.008 **	0.771	0.001 **	0.754	0.001 **
AD_ grasp pen	0.812	0.000 ***	0.760	0.001 **	0.806	0.000 ***	0.815	0.000 ***	0.655	0.011 *	0.680	0.008 **	0.648	0.005 **	0.655	0.008 **

DIS = DIS percentages of DIS tasks; FUNCT = DIS percentages of functional tasks; AD_total = total DIS percentage arm distal DIS tasks; AD_grasp cup = DIS percentage grasp a cup DIS task; AD_grasp pen = DIS percentage grasp a pen DIS task; Total_mean/med = mean/median DIS percentages of all arm distal functional tasks; RF_mean/med = mean/median DIS percentages reach forward tasks; RGV_mean/med = mean/median DIS percentages reach and grasp vertically tasks; RS_mean/med = mean/median DIS percentages reach sideways tasks

***Table 7**: Correlation analyses between DIS score percentages of the DIS tasks and the functional reaching tasks – Choreoathetosis subscale*

FUNCT / DIS	Total_mean		Total_med		RF_mean		RF_med		RGV_mean		RGV_med		RS_mean		RS_med	
	r_s	*p* value	r_s	*p* value	r_s	*p* value	r_s	*p* value	r_s	*p* value	r_s	*p* value	r_s	*p* value	r_s	*p* value
AD_total	0.252	0.365	0.378	0.164	0.234	0.401	0.237	0.395	0.546	0.044 *	0.567	0.034 *	0.226	0.418	0.141	0.617
AD_ grasp cup	0.429	0.110	0.517	0.049 *	0.370	0.175	0.362	0.186	0.561	0.037 *	0.580	0.030 *	0.309	0.263	0.276	0.320
AD_ grasp pen	0.165	0.556	0.280	0.312	0.229	0.411	0.225	0.420	0.518	0.058	0.541	0.046 *	0.116	0.681	0.046	0.870

DIS = DIS percentages of DIS tasks; FUNCT = DIS percentages of functional tasks; AD_total = total DIS percentage arm distal DIS tasks; AD_grasp cup = DIS percentage grasp a cup DIS task; AD_grasp pen = DIS percentage grasp a pen DIS task; Total_mean/med = mean/median DIS percentages of all arm distal functional tasks; RF_mean/med = mean/median DIS percentages reach forward tasks; RGV_mean/med = mean/median DIS percentages reach and grasp vertically tasks; RS_mean/med = mean/median DIS percentages reach sideways tasks

3.2.2 Interrater reliability

The intraclass correlation coefficients (ICC) and 95% confidence intervals (CI) are presented in *table 8*. The duration and amplitude item scores will be discussed in *section 4*.

The DIS score percentages for the distal arm **dystonia subscale** generally showed moderate to good interrater reliability for all functional tasks. A small difference in the mean ICC was found; 0.767 for RS, 0.726 for RGV, and 0.670 for RF. For the DIS dystonia subscale of the RS tasks, ICC values revealed good interrater reliability, ranging from 0.759 to 0.772 (95% CI [0.642-0.838]). Moderate to good reliability coefficients were found for the RGV tasks, ranging from 0.692 to 0.755 (95% CI [0.582-0.825]). ICC statistics in the RF tasks ranged from 0.649 to 0.711 (95% CI [0.503-0.794]), representing a moderate reliability. All 95% CI were significant.

For the **choreoathetosis subscale**, overall lower ICC values were observed for the distal arm DIS scoring of the functional reaching tasks. Mean ICC values for the functional tasks are 0.517, 0.298 and 0.221 for RGV, RS and RF respectively. All DIS scores for the functional tasks showed poor reliability, except RGV1 (ICC=0.671 (95% CI [0.555-0.762])). All 95% CI were significant except RF1 [-0.069-0.227] and RS2 [-0.001-0.325].

Table 8: *Interrater reliability: Intraclass Correlation Coefficients (ICC) with 95% confidence intervals (CI) between raters (k = 2) for the Dyskinesia Impairment Scale scored in functional reaching tasks – Dystonia (DYS) and Choreoathetosis (CA) subscale*

	ICC [95% CI]					
	DYS			**CA**		
	Duration	Amplitude	∑(D+A)	Duration	Amplitude	∑(D+A)
RF1	0.584 [0.436-0.698]	0.634 [0.488-0.740]	0.651 [0.503-0.755]	0.090 [-0.059-0.244]	0.056 [-0.087-0.206]	0.076 [-0.069-0.227]
RF2	0.568 [0.430-0.679]	0.641 [0.504-0.743]	0.649 [0.519-0.747]	0.345 [0.170-0.498]	0.000 [-0.177-0.178]	0.264 [0.164-0.424]
RF3	0.679 [0.561-0.770]	0.628 [0.497-0.731]	0.711 [0.602-0.794]	0.293 [0.113-0.456]	0.324 [0.139-0.486]	0.322 [0.140-0.483]
RGV1	0.687 [0.572-0.775]	0.736 [0.638-0.811]	0.755 [0.663-0.825]	0.678 [0.564-0.767]	0.638 [0.513-0.736]	0.671 [0.555-0.762]
RGV2	0.527 [0.343-0.665]	0.702 [0.595-0.784]	0.692 [0.582-0.777]	0.321 [0.150-0.474]	0.481 [0.329-0.609]	0.410 [0.248-0.550]
RGV3	0.585 [0.414-0.710]	0.712 [0.600-0.797]	0.732 [0.626-0.811]	0.441 [0.247-0.596]	0.480 [0.274-0.636]	0.470 [0.266-0.626]
RS1	0.752 [0.662-0.821]	0.720 [0.586-0.810]	0.770 [0.676-0.838]	0.360 [0.175-0.518]	0.285 [0.113-0.441]	0.344 [0.166-0.499]
RS2	0.685 [0.575-0.770]	0.767 [0.671-0.836]	0.772 [0.687-0.836]	0.149 [-0.015-0.310]	0.146 [-0.020-0.308]	0.165 [-0.001-0.325]
RS3	0.757 [0.658-0.831]	0.693 [0.501-0.806]	0.759 [0.642-0.838]	0.385 [0.154-0.565]	0.375 [0.142-0.557]	0.385 [0.147-0.569]

RF = reach forward; RGV = reach and grasp vertically; RS = reach sideways; Each task was performed three times (1-2-3)

∑(D+A) = summation of the duration and amplitude factors

3.3 Machine learning classifier model

3.3.1 Performance of the multiclass classifier model

Table 9: *Performance measures (%) of the multiclass classifier - total and fine-tuned dataset*

	ACCURACY	F1-MEASURE	PRECISION	RECALL
Total dataset	71.85%	71.85%	71.49%	71.82%
Multiclass fine-tuned dataset	74.58%	74.11%	74.97%	74.58%

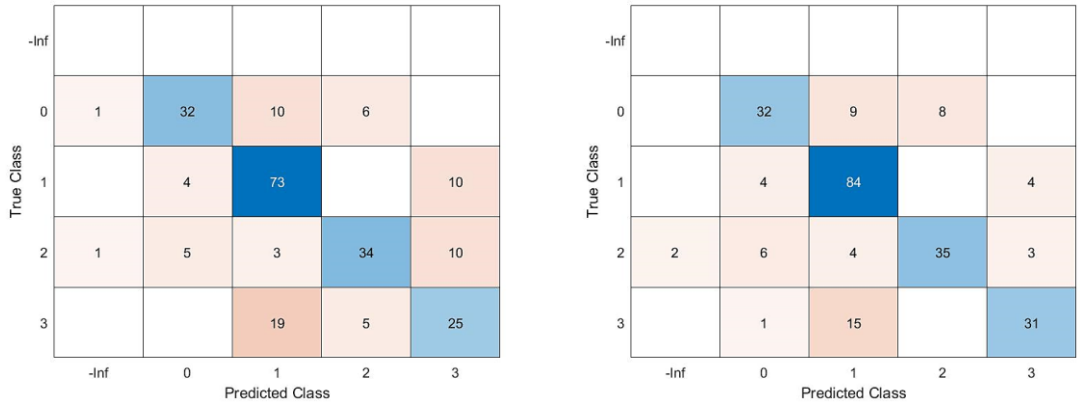


Figure 8: *Confusion matrices of the multiclass classification a) Total dataset ; b) Multiclass fine-tuned dataset*

Feature reduction

The results obtained after running the total dataset in the SVM multiclass (classes 0-3) model are presented in *table 9* and *figure 8*. Performance measures were automatically calculated in MATLAB based on the confusion matrix. An illustration of the confusion matrix working mechanism was given in *section 2.6, table 5.* The performance measures should be interpreted as followed: observing higher performance measures after excluding a single feature is an indication that this feature is less important in discriminating between classes, and vice versa.

Feature reduction was proceeded using different strategies at different levels: categorical exclusion, refining categories (forward and backward feature set selection, and reasoned combined feature set selection) and fine-tuning. This process, aiming for the most optimal feature input, is presented in *figure 9*.

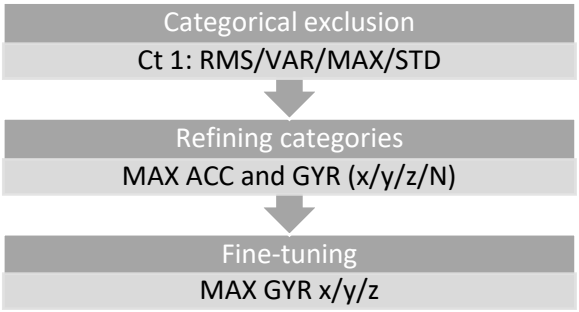


Figure 9: *Flowchart of the feature reduction procedure, illustrated with an example*

A. Categorical exclusion

Features were combined in six categories based on their similarity e.g. RMS, VAR, MAX and STD of acceleration and angular velocity measures were combined in category 1. The importance of the category could be interpreted after exclusion from the total dataset. This was achieved by making a ratio balancing the model performance using the total dataset and the total dataset minus the single excluded category. The results presented in *table 10* should be interpreted as followed: high accuracy when omitting a specific category (e.g. Ct.1 in line 1) from the total dataset proved the inferior importance of the features category.

Table 10: *Performance measures (%) of the multiclass classifier with categorical exclusion (Ct.)*

	ACCURACY	**F1-MEASURE**	**PRECISION**	**RECALL**
Ct. 1: RMS/VAR/MAX/STD	72.06%	72.09%	72.58%	72.06%
Ct. 2: RMSJ/VARJ/MAXJ/STDJ	69.54%	69.02%	69.17%	69.54%
Ct. 3: SE/COR/MAXVEL/DUR	69.33%	69.25%	70.16%	69.33%
Ct. 4: SM/SpE	68.70%	68.45%	69.18%	68.70%
Ct. 5: Power	70.80%	70.78%	71.68%	70.80%
Ct. 6: MOCAP	69.33%	69.19%	70.10%	69.33%
Total dataset	71.85%	71.85%	71.49%	71.82%

Smoothness and spectral entropy (Ct. 4) were found to be more important in the discrimination between the four classes, since exclusion of these features yielded lower model performance. In contrast, acceleration and angular velocity measures (Ct. 1) were considered less discriminative.

B. Refining categories

In this second phase, analysis of features within the six above-mentioned categories was executed. A first step was the leave-one-out feature set reduction based on the total dataset. The feature sets combined the ACC and GYR data from all axes. E.g. the MAX feature set consisted of MAX_ACC and MAX_GYR in x/y/z/N. The results are depicted in *figure 10*. More precise conclusions about feature relevance were feasible in analyzing the performance after exclusion of a single feature set e.g. worse performance with excluding MAX_ACC and MAX_GYR in x/y/z/N (Ct. 1) from the total dataset (accuracy of the total dataset (71.85%) > accuracy of the total dataset minus MAX_AG (70.17%)).

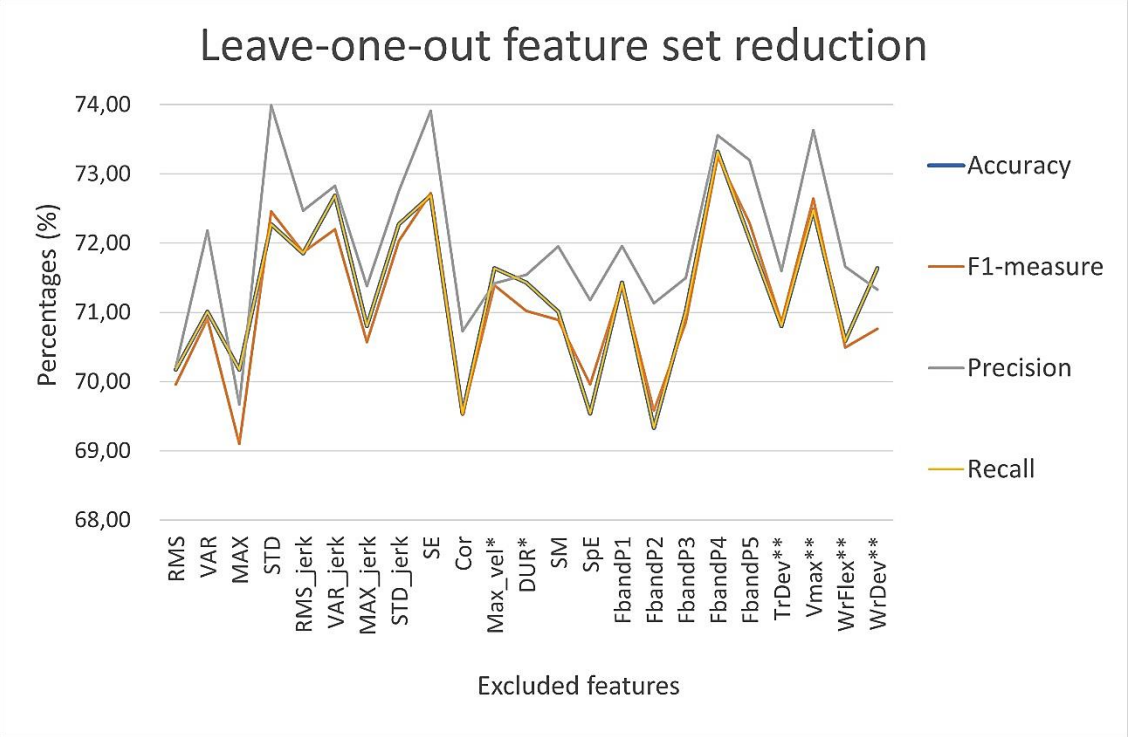


Figure 10: *Performance (%) of the multiclass classifier model with leave-one-out feature set reduction*

Acceleration and gyroscope values for all features except * = only acceleration features; ** = MOCAP features

Leave-one-out feature set reduction showed that maximal acceleration and angular velocity, the correlations between axes, spectral entropy and power in frequency band 0.5-1Hz have an important discriminative capacity, as accuracy significantly decreased when excluding these feature sets from the dataset. Feature sets of less interest based on the leave-one-out analyses were: standard deviation of the acceleration and angular velocity signal, the variance of the linear and angular jerk, sample entropy, power in frequency band 2-3Hz and 3-4Hz, and the maximal linear velocity of the MOCAP system.

The backward and forward feature set selection, and the reasoned combined feature reduction together resulted in the most optimal (non-finetuned) combination of 50 features for the discrimination between the four classes.

C. Fine-tuning

Further fine-tuning of the feature dataset, currently consisting of 50 features from the refining categories phase, was based on the distinction in axes for both accelerometer and gyroscope data. E.g. all features within the feature set MAX_AG: MAX_Ax, MAX_Ay, MAX_Az, MAX_AN, MAX_Gx, MAX_Gy, MAX_Gz and MAX_GN were separately evaluated. Feature sets were fine-tuned in a cumulative way, starting with the fine-tuning of the first selected feature set (MAX_AG), followed by the fine-tuning of the second feature set (RMS_jerk), etc.

Finally, a total of 23 relevant features was deduced from the 144 features in the total dataset. The resulting relevant features for the multiclass model obtained in the cumulative fine-tuning process are listed in *Appendix B, table 1*. After fine-tuning the feature input, an average increase of 3% in the SVM performance measures was noticed relative to the initial total dataset *(table 9),* and a decrease in misclassifications was found for class 1, 2 and 3 *(table 11).*

Table 11: *Misclassification rate (%) in the total and fine-tuned dataset*

	CLASS 0	**CLASS 1**	**CLASS 2**	**CLASS 3**
Total dataset	34.69%	16.09%	35.85%	48.98%
Multiclass fine-tuned dataset	34.69%	8.70%	30.00%	34.04%

3.3.2 Performance of the binary classifier model

Subsequently, an inspection on the relevant features specifically characterizing DYS and CA was performed. The most relevant features in the binary datasets (No DYS/DYS and No CA/CA) were extracted on the basis of leave-one-out and fine-tuning procedures. Additionally, features excluded in the multiclass analysis were added once again to reconfirm that they did not add to increased accuracy values in the binary models.

Feature reduction

A. Multiclass fine-tuned dataset reduction

The multiclass fine-tuned dataset of 23 features was implemented as a starting point for the binary classifier feature reduction process. One by one features were excluded from the dataset. The change in accuracy of both the DYS and CA binary models are presented in *figure 11*.

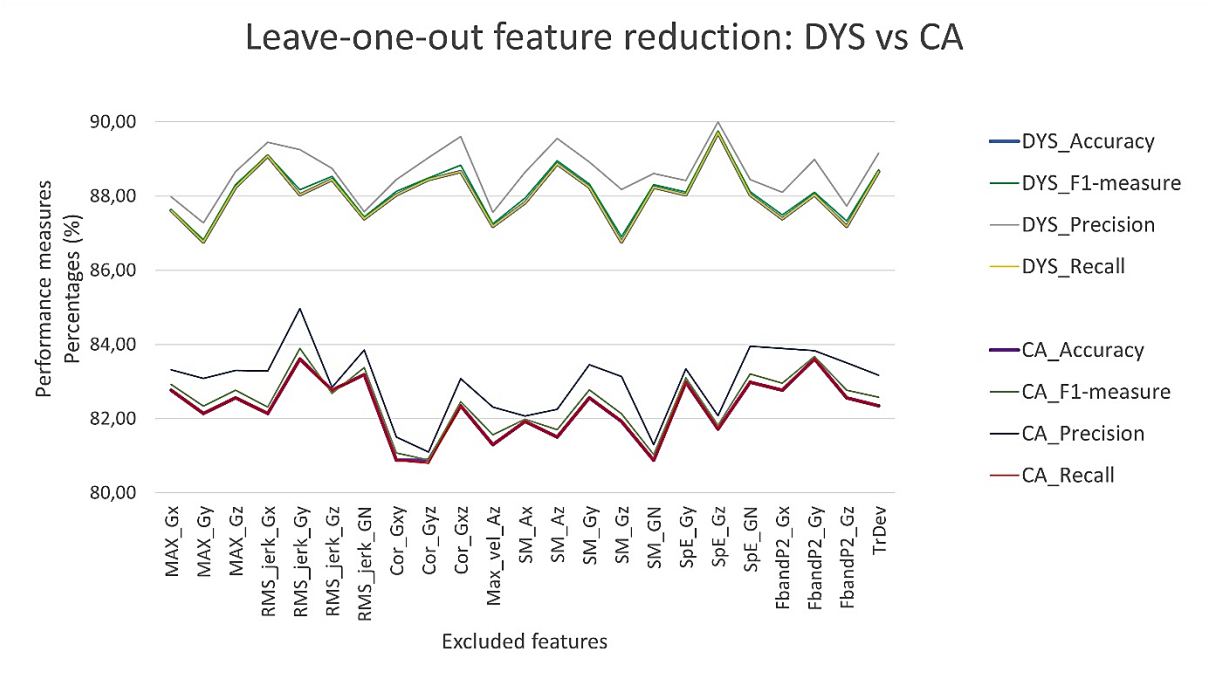


Figure 11: *Leave-one-out feature reduction – change in performance (%): DYS vs. CA dataset*

- BINARY CLASSIFIER MODEL: NO DYS VS. DYS

Table 12: *Performance measures (%) of the DYS binary classifier - multiclass and DYS fine-tuned dataset*

	ACCURACY	F1-MEASURE	PRECISION	RECALL
Multiclass fine-tuned dataset	88.03%	88.12%	88.71%	88.03%
DYS fine-tuned dataset	90.34%	90.35%	90.46%	90.34%

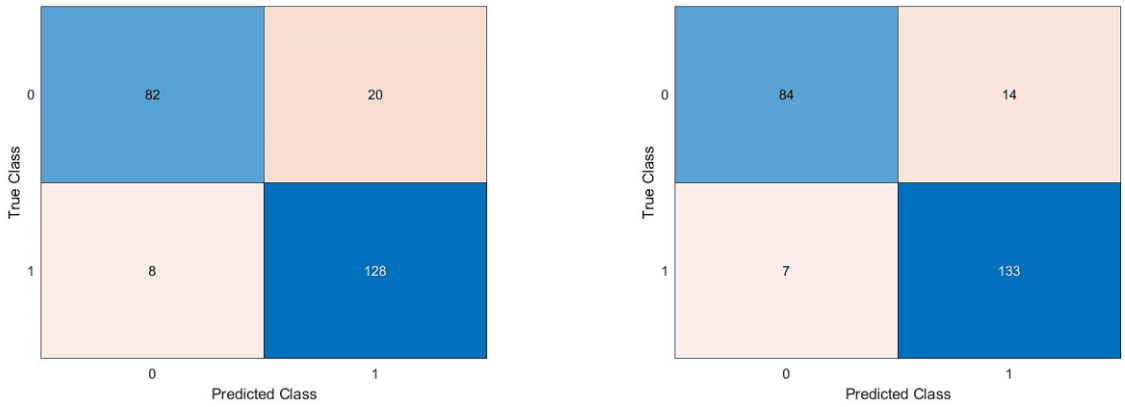


Figure 12: *Confusion matrices of the DYS binary model for the a) Multiclass fine-tuned dataset; b) DYS fine-tuned dataset; with 0 = No DYS and 1 = DYS*

The results obtained after running the multiclass obtained feature set in the DYS binary SVM model and the associated confusion matrices are presented in *table 12* and *figure 12.*

The consequent leave-one-out feature analysis (*figure 11*) on the multiclass fine-tuned dataset demonstrated an important discriminative capacity for the maximal angular velocity – x- and y-axes ; the root mean square of angular jerk – norm value ; linear maximal velocity – z-axis ; smoothness of the gyroscope signal – z-axis ; power of gyroscope signal in frequency band 0.5-1Hz – x- and z-axes.

After submitting the initially excluded feature sets of the total multiclass dataset, the following feature sets showed additional discriminative capacity: variance of acceleration and angular velocity, standard deviation of jerk, power of the accelerometer signal in frequency band 0-0.5Hz and 2-3Hz, and wrist flexion/extension. These feature sets were added to the above-mentioned discriminative features and were all further fine-tuned in the following phase, i.e. distinction in axes for both ACC and GYR data.

- BINARY CLASSIFIER MODEL: NO CA VS. CA

Table 13: *Performance measures (%) of the CA binary classifier - multiclass and CA fine-tuned dataset*

	ACCURACY	**F1-MEASURE**	**PRECISION**	**RECALL**
Multiclass fine-tuned dataset	82.77%	82.94%	83.50%	82.77%
CA fine-tuned dataset	84.66%	84.79%	85.13%	84.66%

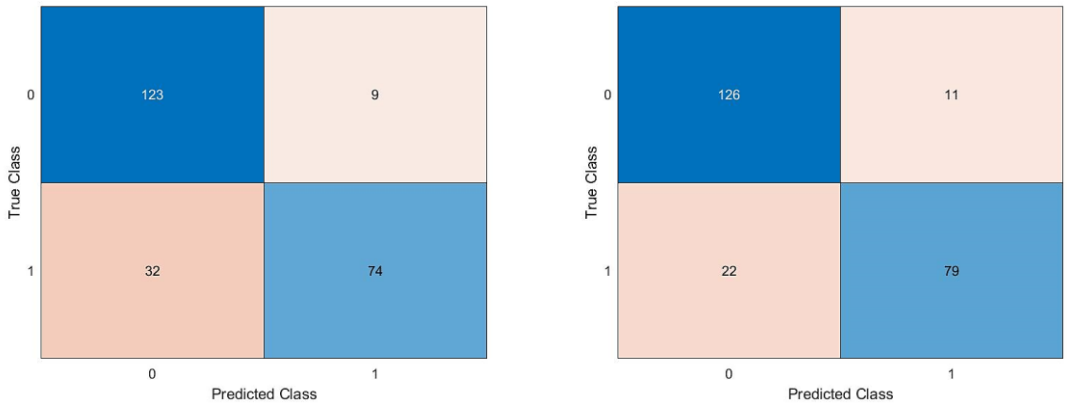


Figure 13: *Confusion matrices of the CA binary model for the a) Multiclass fine-tuned dataset; b) CA fine-tuned dataset; with 0 = No CA and 1 = CA*

The results obtained after running the multiclass obtained feature set in the CA binary SVM model and the associated confusion matrices are presented in *table 13* and *figure 13.*

The consequent leave-one-out feature analysis (*figure 11*) on the multiclass fine-tuned dataset demonstrated an important discriminative capacity for correlation of the x/y- and y/z-axes of the gyroscope signal ; linear maximal velocity – z-axis ; smoothness of the accelerometer signal – x- and z-axes ; smoothness of the gyroscope signal – z-axis and norm value ; spectral entropy of the gyroscope signal – z-axis.

After submitting the initially excluded feature sets of the total multiclass dataset, only wrist deviation angles showed additional discriminative capacity. This feature set was added to the above-mentioned discriminative features and were all further fine-tuned in the following phase, i.e. distinction in axes for both ACC and GYR data.

B. Fine-tuning

A similar method as used in the multiclass dataset fine-tuning process was executed; fine-tuning of the feature dataset was based on specification of axes for both acceleration and gyroscope data.

- BINARY CLASSIFIER MODEL: NO DYS VS. DYS

A total of 22 features was derived after feature reduction and fine-tuning of the DYS dataset. These features are listed in *Appendix B, table 1.* The performance of this binary classifier model and the percentage of misclassifications is presented respectively in *table 12 and 14.* A change of 2% in performance was observed comparing the multiclass fine-tuned dataset and the ultimate DYS fine-tuned dataset. In general, less misclassifications were found in the DYS class (class 1).

Table 14: *Misclassification rate (%) in the multiclass fine-tuned and DYS fine-tuned dataset*

	CLASS 0 (No DYS)	**CLASS 1 (DYS)**
Multiclass fine-tuned dataset	19.61%	5.89%
Fine-tuned DYS dataset	14.29%	5.00%

- BINARY CLASSIFIER MODEL: NO CA VS. CA

A total of 21 features was derived after feature reduction and fine-tuning of the CA dataset. These features are listed in *Appendix B, table 1*. The performance of this binary classifier model and the percentage of misclassifications is presented respectively in *table 13 and 15.* A change of 2% in performance was observed comparing the multiclass fine-tuned dataset and the ultimate CA fine-tuned dataset. In general, more misclassifications were found in the CA class (class 1).

Table 15: *Misclassification rate (%) in the multiclass fine-tuned and CA fine-tuned dataset*

	CLASS 0 (No CA)	**CLASS 1 (CA)**
Multiclass fine-tuned dataset	6.82%	30.19%
Fine-tuned CA dataset	8.03%	21.78%

- COMPARISON DYS VS. CA

The feature distribution of the fine-tuned dataset for both DYS and CA is presented in *figure 14.* The y-axis represents the number of features (number of axes: x, y, z, N) that were of interest. This comparison demonstrated that variance and maximal angular velocity, root mean square of the angular jerk, standard deviation of the linear jerk, the correlations between axes of the gyroscope, smoothness of the accelerometer data, power in frequency band 0-0.5Hz and 2-3Hz of the accelerometer data and wrist flexion/extension angles could clearly describe the dystonic movements. Characteristic features found for describing the choreoathetotic movements were maximal linear velocity of the accelerometer signal, smoothness and power in frequency band 0.5-1Hz of the gyroscope signal, and wrist deviation angles.

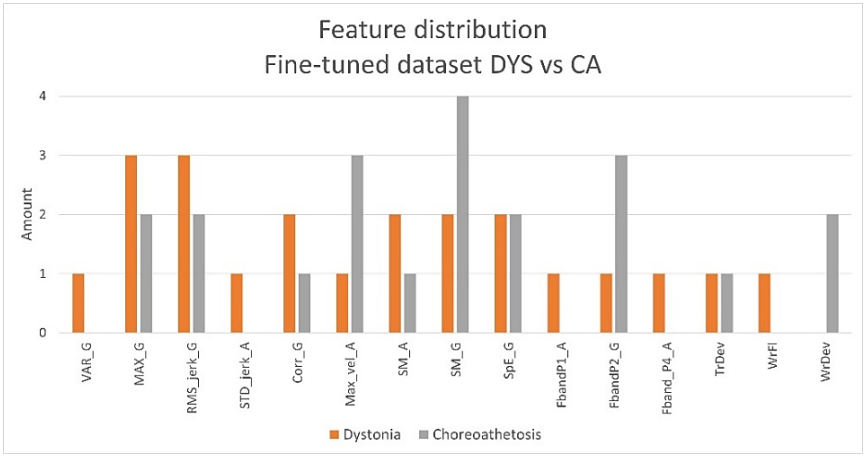


Figure 14: *Relevant feature distribution based on the feature type in DYS and CA fine-tuned datasets*

The relevance of acceleration, gyroscope and motion capture data is depicted in *figure 15*. In both DYS and CA, a higher representation of gyroscope features could be found for the fine-tuned datasets. Acceleration features were more important in the representation of dystonic movements. In contrast, the MOCAP features were more important to describe choreoathetotic movements.

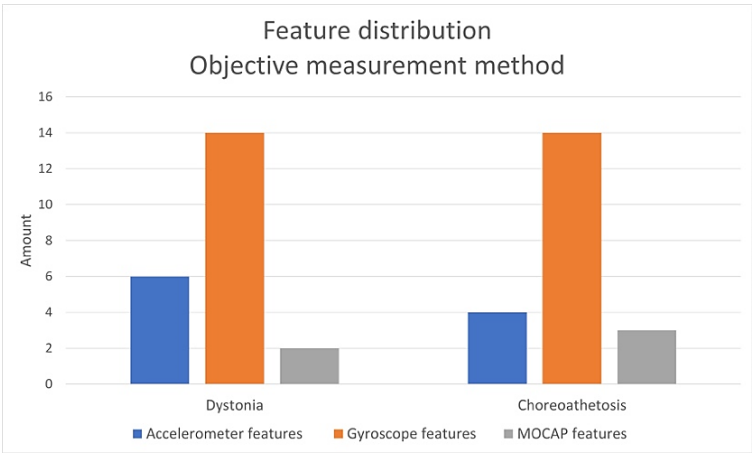


Figure 15: *Relevant feature distribution based on the measurement methods for the DYS and CA fine-tuned datasets*

4. Discussion

Previous studies within the DCP population indicated the benefit of using objective measurement tools for evaluating and discriminating DYS and CA.[48] However, no study yet focused on the minority of children within the DCP population who are able to perform functional upper limb tasks.
Accurate discrimination of DYS and CA is important for improving insights, correct treatment and quality of life in the DCP population. The ultimate aim of this study was to automatically discriminate DYS and CA using data from both MOCAP and IMU in a classifier model. In addition, the most relevant features characterizing each motor disorder were identified. As a sub-aim, this study investigated the validity and reliability of the DIS used in the ULEMA protocol.

4.1 Concurrent validity

In general, a difference in tasks between the ULEMA protocol and the original DIS tasks did not have a significant effect on the DIS severity scores for **dystonia**. The results showed moderate to excellent associations for the entire dystonia subscale, with the strongest correlations for the functional RF tasks and the DIS grasp-a-pen task. This could be attributed to the equalities between both tasks i.e. a static wrist and forearm position.

A previous study of Monbaliu et al. compared the DIS dystonia subscale with the BADS scale. Pearson's correlation coefficient was 0.84, revealing an excellent relationship.[27] This is in line with the calculated mean of the results found in this study, namely a Spearman correlation of 0.78.

In contrast with the dystonia subscale, the correlations in the **choreoathetosis subscale** were absent to moderate. Only the RGV tasks were correlated with the DIS tasks. This could be attributed to the difficulty of the tasks. DIS tasks were generally perceived more difficult, resulting in higher score percentages. The RGV tasks were more challenging with respect to coordination in the distal arm compared to the other functional reaching tasks. The comparability of the RGV tasks with the DIS tasks could lead to comparable severity percentages, possibly evoking better validity.
Future research should investigate the relative function of all three functional reaching tasks in representing the dystonic and choreoathetotic movements in the machine learning process.

Choreoathetosis is characterized by variable chaotic movements, which could impede uniform scoring, especially for the movement amplitude. Moreover, dystonia is more prominently and more frequently present which could lead to lower noticeability of choreoathetosis, making it harder to score the latter.[12] The writhing fingers movements were typical signs of choreoathetosis in the hand, but the fingers were excluded from the analyses of both scoring situations. However, these finger movements

were a disturbing factor during the scoring of the DIS of the functional reaching tasks as it was difficult not to take them into account. Another difficulty during the scoring was the dubiety about the origin of the choreoathetotic movements (proximal or distal) as well as the doubt about the occasionally vague division between a normal reaction and an abnormal movement e.g. the recoil at point of task achievement.

Comparison with other studies could not be made, since this was the first study to investigate the concurrent validity of the clinical scoring of choreoathetosis.

Striking, focusing on the concurrent validity of the objective measurement of DYS and CA, Haberfehlner et al. indicated a better relation with choreoathetosis. The study did not point significant relations for the objective measurement and the DIS dystonia subscale. Though, it should be noted that in this study involuntary movements in the lower limb were analyzed in a resting position and in participants with a GMFCS level IV and V.[61]

Some important aspects need to be considered with respect to the difference between both DIS applications. A first consideration is the discrepancy of the task complexity, which could be noticed in lower score percentages for the functional reaching tasks after examining the raw data. Another aspect to take into account should be the difference in the scoring formula; the DIS scoring of the functional reaching tasks was based on the mathematical mean of the eight repetitions separately, whereas the DIS scoring of the DIS tasks took place by scoring ten repetitions all at once. The latter implicates that only the highest severity presentation was reflected in the DIS scoring. A third consideration lies in rescaling the duration item score from a 5-point to a 3-point scale in scoring the functional tasks, which implies less gradation in possibilities of score percentages. E.g. DIS duration item score 3 is interpreted as a 75% severity score for the motor disorder, whereas 75% is impossible to score with the DIS in the functional reaching tasks i.e. 0%, 50% and 100% are the only score possibilities. The DIS duration item score 3 immediately reflects in a 100% severity score in the DIS in the functional reaching tasks No conclusions could be made based on further analysis on the duration and amplitude item scoring in both DIS and functional reaching tasks.

4.2 Interrater reliability

For the **dystonia subscale**, the DIS severity percentages showed moderate to good interrater reliability for the functional reaching tasks, with the best results for the functional RS tasks. In general, the reliability scores of dystonia were fairly in line with the excellent interrater reliability scores found in the DIS reliability studies on experienced and unexperienced raters.[27, 62] In the ICC of the item scores, better results were generally found for amplitude.

For the **choreoathetosis subscale** however, poor ICC values were found for the DIS severity score, except for RGV1. The more conform scoring of choreoathetosis in the RGV tasks was also recognized in the concurrent validity results. The difficulties experienced during the DIS scoring process and the variable presence and expression of choreoathetosis as previously mentioned could be explanations for the poor ICC results. In contrast with the dystonia subscale, ICC results of the choreoathetosis subscale were lower compared to previous studies.[27, 62]

Interrater differences in the original DIS scoring were comparable with the results found for both experienced and unexperienced raters in previous studies.[27, 62] Thus, the original DIS scoring was a reliable input in the concurrent validity study.

4.3 Machine learning classifier model: DIS classification in functional reaching tasks

Since classifications were needed to compare input (MOCAP and IMU features) and output (DYS and CA classes) of the SVM model, reliable DIS classification of DYS and CA in the functional reaching tasks was required. However, poor results for interrater reliability on the DIS classifications were found (*Appendix A, table 2*). Reviewing the raw data, interrater differences in class 0 (No DYS/No CA) and 2 (No DYS/CA) were highlighted more specifically, pointing towards the role of CA. These findings are visualized in *figure 16.* Previous research already indicated that choreoathetosis was consistently scored lower than dystonia.[12](personal communication) The subtle changes contribute to the difficulty to detect the presence of this motor disorder. This indicated once more that choreoathetosis is more difficult to score, reinforcing the findings described in *section 4.1*. Therefore, results obtained in the SVM for choreoathetosis (*section 3.3*) should be treated with caution.

As mentioned in the methodology, the initial scores of both rater 1 and rater 2 were used in the ICC statistical analyses. After a revision of the scores, the consensus scores were included in the further course of this study. For this reason, the reliability of the classifications included in the SVM model can be justified.

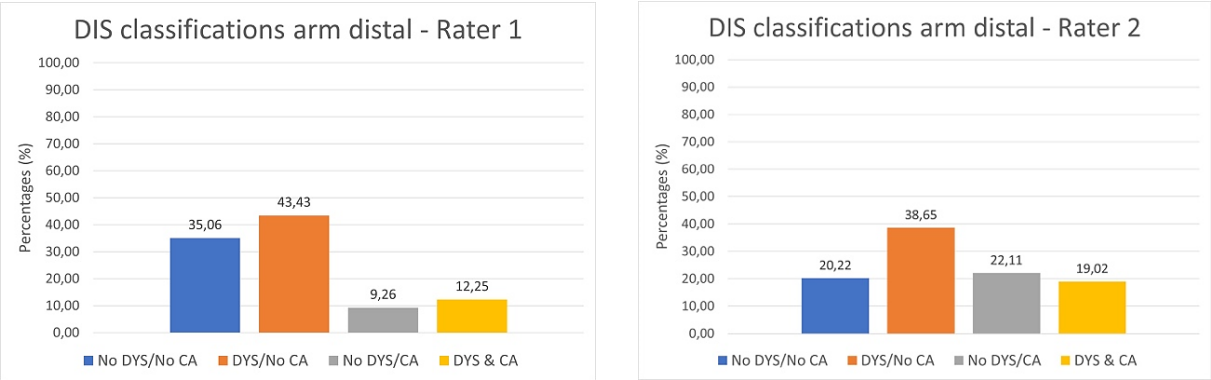


Figure 16: *Comparison of DIS classifications in the distal arm for rater 1 and rater 2*

4.4 Machine learning classifier model: feature extraction and discriminative validity

4.4.1 Multiclass classifier model

This was the first study to develop and test machine learning models for the automated discrimination of DYS and CA in children with DCP during functional upper limb tasks. This study is still an explorative study and must therefore be interpreted on this basis. The SVM algorithm using the final dataset of 23 features was able to classify the four classes with performance measures of approximately 74.5%, and a misclassification rate below 35%. The performance outcomes of the multiclass model were in line compared with the existing studies in literature regarding the determination of movement patterns (*table 16A*).[63-66] Less misclassifications were noted in class 1 (DYS/No CA), which could be explained by the imbalanced class distribution (i.e. 355/948 observations in class 1) and the more reliable and valid scoring of dystonia. In comparison with previously used machine learning classifiers in clinical applications such as SVM, random forest and decision trees (*table 16B*), the performance of this SVM classifier was similar.[63-66] Logistic regression was found to be less accurate as a part of multiclass classification problems.[65, 66]

Table 16A: *Comparison of the SVM performance in the proposed multiclass algorithm to existing multiclass algorithm models in literature*

	ACCURACY	**ERROR RATE**	**F1-MEAS**	**PRECISION**	**RECALL**
Goodlich, B (2020)	77.3%	-	74.1%	-	-
Zhang, Y (2019)	85.0%	5.7%	-	-	-
Butt, A (2018)	74.1%	-	-	-	41.7%
Bidabadi, S (2019)	62.3%	-	-	-	-
Obtained SVM results (section 3.3.1)	**74.58%**	**8.70-34.69%**	**74.11%**	**74.97%**	**74.58%**

Table 16B: *Comparison of the performance of different machine learning algorithms in literature*

	ACCURACY	ERROR RATE	F1-MEAS	PRECISION	RECALL
Goodlich, B (2020)					
- Random forest	79.0%	-	76.6%	-	-
- Decision tree	76.5%	-	72.8%	-	-
Zhang, Y (2019)					
- Random forest	83.6%	6.4%	-	-	-
- Decision tree	84.3%	5.7%	-	-	-
Butt, A (2018)					
-Logistic Regression	70.4%	-	-	-	58.3%
Bidabadi, S (2019)					
-Logistic Regression	54.2%	-	-	-	-
-Random forest	67.4%	-	-	-	-

Apart from the class distribution, the impact of classification selection on the multiclass classifier model should be considered in interpreting the above-mentioned results. As classes were created based on the absence or presence of the motor disorder, a large variation in the presentation of severity was recognized in class 1, 2 and 3. This complicated the detection of clear patterns during the training phase of the SVM model, especially in classes with smaller sample sizes.

For the initial study, a linear regression model using continuous data was not possible due to the large number of included features. Future research could be done using the obtained 23 features of the multiclass model from this study in a multiple linear regression model, aiming to find a linear relationship in the input and output variables of a given dataset. The interpretability of this linear regression model is found superior, caused by the absence of black box methodology. Moreover, the linear regression is a computationally more efficient method. Nonetheless, a lower performance accuracy is found as compared to other regression models. The linear regression machine learning model is less experienced in dealing with a high amount of data and unbalanced classes, and shows a higher amount of misclassifications.[67] Previous studies showed that this higher amount of misclassifications is remarkable in dystonia.[48] Specifically, this model is sensitive for outliers and the presence of underfitting, caused by the typically more complex nature of data.[67] Therefore, also the more complex and less efficient non-parametric regression models should be considered in future research. Comparing the performance of different machine learning models, SVM and gaussian process regression were indicated as better regression models for quantifying dystonia, whereas linear regression and random forest regression were better for choreoathetosis.[48]

4.4.2 Binary classifier models

DYS and CA binary classifiers were important to improve knowledge of characteristics for each motor disorder. As binary classification is a less complex function, better performance outcomes were found regarding the classification of No DYS/DYS and No CA/CA. The results for both motor disorders were in line with similar studies on the SVM performance, as presented in *table 17A*.[55, 68-70] *Table 17B* represents the comparison of the performance of different classifier models based on literature.[68-70]

Table 17A: *Comparison of the SVM performance in the proposed binary algorithm to existing binary algorithm models in literature*

	ACCURACY	**ERROR RATE**	**F1-MEAS**	**PRECISION**	**RECALL**
Kim, JY (2020): Spastic CP	71.8%	-	-	-	-
Cole, B (2014): Tremor	-	7.2%	-	-	-
Cole, B (2014): Dyskinesia	-	9.1%	-	-	-
Bennasar, M (2018): Huntington	91.1%	-	-	-	90.4%
Obtained SVM results DYS (section 3.3.2)	**90.34%**	**5.0-14.29%**	**90.35%**	**90.46%**	**90.34%**
Obtained SVM results CA (section 3.3.2)	**84.66%**	**8.03-21.78%**	**84.79%**	**85.13%**	**84.66%**

Table 17B: *Comparison of the performance of different machine learning algorithms in literature*

	ACCURACY	**ERROR RATE**	**F1-MEAS**	**PRECISION**	**RECALL**
Kim, JY (2020): Spastic CP					
- Decision tree	76.6%	-	-	-	-
- Random forest	91.8%	-	-	-	-
- Linear discriminant analysis	80.6%	-	-	-	-
Cole, B (2014): Tremor					
-Diffuse Neural Network	-	6.2%	-	-	-
Cole, B (2014): Dyskinesia					
-Diffuse Neural Network	-	8.7%	-	-	-
Nica, I (2019): DYS					
- Random forest	80.9%	-	-	-	-
Nica, I (2019): CA					
- Random forest	70.0%	-	-	-	-

A clear overview of the obtained relevant features of each motor disorder and their performance plots is given in *Appendix B, table 1* and *figure 1*. As a clarification, *figure 17* illustrates the defined coordinate system for the accelerometer and gyroscope.

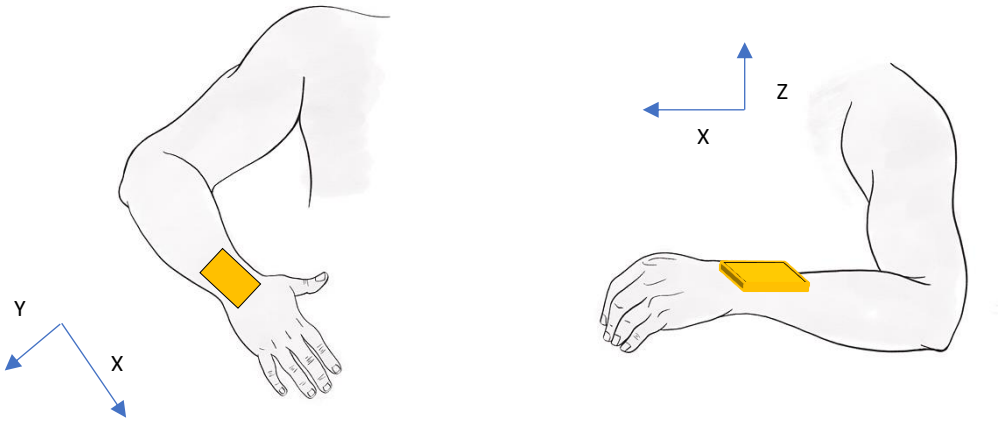


***Figure 17**: Illustration of the coordinate system of the forearm arm IMU in the a) transverse plane and ; b) sagittal plane*

Twenty-two features were of interest in characterizing **dystonia**; these will be described below. Regarding movement axes, movements around and along the z-axis appeared to be most discriminative for No DYS / DYS.

Hypotheses based on literature (*section 2.5.2*) were confirmed in the results of this study regarding angular velocity, correlations between axes, duration, the power in lower frequency bands and wrist flexion/extension.

A continuous alternation of dealing with the tendency to extreme dystonic postures in controlling functional reaching tasks was recognized. In the distal arm, the dystonic postures emerged as a significantly flexed wrist position.

The dystonic movements were mostly three-dimensional, partly objectified by strong correlations between the coordinate axes for the gyroscope signal. Movements around the y-axis often concurred with movements around the x- or z-axis, i.e. elbow flexion, forearm pronation and external shoulder rotation, deviating towards the upper limb flexion pattern. Again, this represented the less coordinated, less straight reaching, discussed as a characteristic of dystonia. Less coordinated movements were previously suggested to be associated with excessive antagonist muscle activation and overflow activation of non-essential muscles.[46, 71-74]

The presence of movement fragmentation, velocity resistance and the efforts to control the performance and precision of sequenced movements could have resulted in the high movement duration.[33, 46, 73, 75, 76]

Results of this study indicated that dystonia was related to a slightly lower maximum and variance of the angular velocity, respectively in three dimensions and around the z-axis. Low variance was not expected but could possibly be explained by a decrease in degree of freedom due to the tendency towards a sustained upper limb flexion pattern in more severe hypertonia. However, an increased movement variability in dystonia was frequently reported in previous studies.[46, 52, 73, 77] The higher presence of involuntary movements in the dystonia group accompanied the resulting inability to fully control motor actions and stabilize joints.[46, 73, 76-79] The more significant stuttering reaching movements and the attempts to restore motor control were expected in children with less severe hypertonia, and could be reflected in repetitive interrupting rotational movements causing variety in acceleration. Focusing on the degree of the angular velocity, dystonic movements were expected to be slow in order to enhance motor performance and target precision, and to avoid an additional increase of movement variability.[73, 75, 76, 78, 80] These findings were confirmed with slightly lower maximal angular velocities in the DYS class with respect to the No DYS class, represented in *Appendix B, figure 1*.

Slightly lower jerk values were hypothesized for dystonia and reflected in the feature plots. (*Appendix B, figure 1*) This should not be interpreted as smooth movements in the DYS group, but rather as more smooth or less unsmooth movements with respect to the No DYS group. The smoother nature of the prominently rotating movements in dystonia could be induced by the presence of class 2 (No DYS/CA) in the No DYS class. In contrast, significantly higher jerk values did emerge in previous studies in the DCP population during analyses in a state of relative rest and in reaching.[46, 48] More negative spectral arch length (smoothness) values were found to be discriminative, representing the unsmooth nature of dystonia. This is in contrast with the previously discussed results of lower jerk values. As past research pinpointed spectral arc length to be the most sensitive measure for evaluating smoothness, the interpretation of the unsmooth nature of dystonic movements could be supported.[51, 58] Clear lower values of spectral entropy were reflected in the feature plots and represented a lower complexity of the dystonic movements.

The arrhythmicity of dystonic movements was clear from the more broad and flat power spectrum graph for dystonia with respect to no dystonia, represented in *Appendix B, figure 2*. Hence, the time series of the reaching movements were composed of a large variety of frequencies. In general, a lower power was noticed in the DYS class with respect to the No DYS class, possibly explained by the presence of the class 2 (No DYS/CA) in the No DYS class. Power of the accelerometer and gyroscope signal was found higher in the lower frequency bands, as hypothesized for dystonia. Focusing on the accelerometer signal, power signals in frequency band 2-3Hz were also found to be discriminative, which was not anticipated.[48] Though, this finding was advocated in one study by the presence of

higher frequencies (4-10Hz) analyzing the dystonic tremor in Parkinson patients.[81] A second study indicated dystonia in the DCP population to be related to the 1.7-3.5Hz frequency range.(personal communication) However, it should be noted that the discriminative ability found in this study lay within a lower power in these higher frequency bands, as presented in *Appendix B, figure 1*.

Prediction error rates were found to be sufficient when focusing on class 1 (DYS) in the DYS binary classifier. However, a high number of misclassifications could be discerned regarding class 0 (No DYS). Results of these findings were listed in *section 3.3.2*, *table 14*. The better performance in detecting DYS could be explained by the high number of observations in class 1 (539 of the 948 observations) when training this class. Larger datasets with more variability are vital in future research.
A second reason for the high number of misclassifications in class 0 concerned the fact that the DIS classifications were solely based on the observed movements in the distal arm region. However, as movements in the proximal arm could have an impact on the data collected by the objective measurements, this could have imposed misclassifications in class 0 for dystonia.

Twenty-one features were of interest in characterizing **choreoathetosis**. In general, movements around the x-axis, translating into the presence of pronation/supination movements, and norm values of the features emerged.

Hypotheses based on literature (*section 2.5.2*) were confirmed in the results of this study regarding angular jerk, correlations between axes, linear and angular smoothness values, linear and angular spectral entropy and trajectory deviation.
First, the high amount of change in angular acceleration around the z-axis and its norm value strongly related to noisy, unsmooth, chaotic movements. Specifically, chaos could be noticed in a continuous change in direction and speed during shoulder rotating movements and/or elbow flexion/extension in the horizontal plane. These less coordinated movements usually arose at the same time with movements into forearm pronation/supination, i.e. around the x-axis.
The concomitant presence of involuntary and less coordinated movements did not always translate into an increased movement duration. This finding was in line with previous research.[82]
The discriminative capacity of trajectory deviation could be translated in excessive and more deviations from the normal pathway in choreoathetotic movements.

Subsequently, linear and angular spectral arc length (smoothness) turned out to be dominant parameters, with less coordinated movements for the CA class. Analysis of the smoothness plots (*Appendix B, figure 1*) generally represented more negative values for the norm of the translational movements and the three-dimensional rotational movements in this class. In past research, spectral

arc length was pinpointed as the most sensitive measure for evaluating smoothness, and was highlighted as a distinguishing measure of cerebral palsy and the healthy population.[51, 58]
The presence of unpredictable complex movements in choreoathetosis was confirmed in the SVM frequency outcome results by the spectral entropy of the gyroscope signal. Validation of these findings was given in research on chorea in Huntington and the DCP population, and on Parkinson's tremor.[48, 55, 83] However, when interpreting the feature plots (*Appendix B, figure 1*), no clear differences could be determined. Lower values were found to be representative for dystonia, reflected in class 1 and 3 (DYS/ No CA and DYS & CA). The remaining classes 0 (No DYS/No CA) and 2 (No DYS/CA) showed higher spectral entropy values, indicating more complex movements. Since both higher and lower values were present in both No CA and CA classes, no defined conclusions could be made about the movement complexity.

A more interesting and unexpected finding in choreoathetosis was the presence of higher linear velocities in 3D for the accelerometer axes. This could be related to the continuously rapid changing movement directions, characterizing hyperkinesia.[71]
As a second contrasting finding, the natural position in children with choreoathetosis was found to be oriented towards ulnar deviation.
Entropy measures were expected to characterize choreoathetosis. It should be noted that this was not the case in this study for sample entropy, as previously anticipated by Vanmechelen et al. for the gyroscope signal data of the distal arm.[84]
In general, the more spiked structure of the power spectrum in the CA class with respect to No CA class should be noted in frequency band 0.5-1Hz. A similar gradient of the power in higher frequency bands was demonstrated, represented in *Appendix B, figure 2*. Previous studies reported a tendency towards a repetitive pattern in dyskinesia, which could be deducted from this consistent peak in the power spectrum in the lower frequencies.[85-87] The peak was reflected in clearly higher power of the gyroscope signal localized in frequency band 0.5-1Hz, which is in contrast with the expected significant presence of choreoathetosis in the higher frequency bands. Notwithstanding the ambiguity in literature about this finding, Cuyvers et al. reported a range in frequencies of 0.8-3.5Hz; Burkhard et al. documented a peak frequency at 1.5-3.25Hz.[48, 87] A possible explanation of the absent discriminative value of power in higher frequency bands could be the inclusion criteria of MACS level I-III, meaning that children with an insufficient manual function were excluded from this study. Including more severe cases of choreoathetosis, in an adapted protocol, could facilitate the SVM learning process in future research and could improve the current insights for choreoathetosis.

The choreoathetosis results should be interpreted with caution. In the initial analyses, the results of the concurrent validity and interrater reliability study were insufficient for choreoathetosis. Moreover, a clear imbalance in the DIS classification distribution emerged, with a higher presence of training data for class 1 (DYS/No CA) in the multiclass model. This could be translated in a higher presence of training data (558 of the 948 observations), a better performance and less misclassifications for class 0 (No CA). Although high classification performance was achieved in total, this could not ensure the capability of the algorithm to provide the same results in equal sample sizes for both classes. More specifically, a high number of misclassifications in class 1 (CA), i.e., CA classified by the raters, but not predicted by the binary model, partly restrained the generalization of the results.

For both DYS and CA, translational movements were less representative compared to the rotational movements, illustrated in *figure 15*. The relevance of gyroscope data could be explained by the largely rotational nature of human movements.[88] The power spectrums in *figure 2* in *Appendix B*, represented this more important discriminative value of the gyroscope data, with a larger discrepancy between both motor disorders i.e. larger distance between the curves, with respect to the accelerometer data. In general, accelerometer signals were found to be of more interest in dystonia.

Notwithstanding their similarities, the results obtained in this study highlighted the separate entity of both motor disorders. Unique characteristics for dystonia with respect to choreoathetosis were the variance of the angular velocity, standard deviation of the linear jerk, power in frequency band 0-0.5Hz and 2-3Hz of the accelerometer signal, and the minimal wrist flexion/extension angles. Optimal description of choreoathetosis with respect to dystonia was possible using discriminating features such as the maximal linear velocity, smoothness of the gyroscope signal, power in frequency band 0.5-1Hz of the gyroscope signal, and minimal/maximal wrist deviation angles.

Time domain measures were of higher interest in classification, with a slightly higher importance for dystonia (59.1% of the total features) compared to choreoathetosis (52.4% of the total features). The consequent more important role of frequency measures in CA with respect to DYS is in line with results of previous research.(personal communication)

Notwithstanding the existing ambiguity, more significant differences were expected between both motor disorders regarding the power spectrum. In contrast to some previous results, in this study no significantly higher power was found for choreoathetosis in the higher frequency bands.[48, 56, 87] Burkhard et al. described dyskinesia in Parkinson's disease pointing out a broad power range with a predominant frequency from 0.25 to 1.25Hz and 1.5 to 3.25Hz for dystonic and choreiform dyskinesia respectively.[87] Cuyvers et al. studied the DCP population in a state of relative rest and encountered frequencies ranging from 0 to 0.8Hz for dystonia and 0.8 to 3.5Hz for choreoathetosis.[48]

Discriminative ability of power in the higher frequency bands was found for dystonia with respect to choreoathetosis. A lower power in dystonia in these higher frequencies was already mentioned above.

As noted, results were not always anticipated and reflected in the feature plots. Three general conclusions could be made based on these feature plots. First, choreoathetosis was reflected in highly unsmooth movements with respect to dystonia. Towards a second conclusion, a more spiked and energetic power spectrum was revealed in choreoathetosis compared to dystonia. These curves suggested dystonic movements to be more arrhythmic. A stronger rhythmicity could be found in choreoathetosis. At last, dystonic movements were found to be less complex, whereas in choreoathetosis no conclusions could be made.

In the previous discussion, frequent discrepancies existed regarding the obtained relevant features and the concomitant feature plot interpretation. As SVM uses black box approach in the automatic classification, the process used to obtain the most relevant feature set could not be determined.

4.5 Limitations

This study had some limitations and therefore warrants some critical reflections. First, only 12 children were included. In the future, larger sample sizes are needed to allow for more generalized conclusions regarding the reliability and validity of the DIS in the functional tasks. Still, a sufficient number of observations were obtained to allow sufficient SVM power. A more equal distribution over different severity levels and classes is demanded. Furthermore, there was a restriction in inclusion criteria. Due to the choice of functional reaching tasks in the ULEMA protocol, patients with DCP and a MACS level IV or V were excluded. However, studies indicated that about 65% of the children with DCP are classified in the severe levels of manual ability.[11] In this less manually abled group of children with DCP, research on the use of IMU devices in discriminating DYS and CA had already been conducted.[48] Since the aim of this study was to objectively discriminate DYS and CA during the functional tasks, the participants had to be able to execute the requested reaching and grasping tasks. Future research should include all different levels of manual ability, aiming for a better generalization and performance of the SVM.

Another future perspective is the evaluation of this model in the context of activities of daily life, which could be feasible if only including IMU features. Both IMU and MOCAP systems were used to evaluate the kinematics of the functional tasks. Both systems were combined because of their complementarity, leading to an increased chance to obtain the most relevant set of features. The limitations of the optometric system however lie within the time-consuming aspect of both placement and labelling of the markers. Moreover, a fully equipped laboratory, expertise in palpation and data processing is

needed. Evaluation was done on the ability to eliminate the use of MOCAP in the protocol to obtain a time- and cost-efficient, portable classifying system. However, results clarified the value of MOCAP in the discrimination of dystonia and choreoathetosis.
This study was limited to the inclusion of the distal arm data. Only the wrist and forearm data were implemented, respectively obtained by MOCAP and IMU. Upcoming analyses should explore the impact of including the entire upper limb. However, this requires a large number of participants to preserve the statistical power of the SVM model.

Ultimately, the presence of spasticity in some children with DCP should also be taken into account when interpreting the results. This could potentially have an influence on the smoothness, range of motion and velocity of the upper limb movement pattern. Movement velocity is by nature complicated to control in children with dyskinesia, which has an impact on the severity of spasticity known to be velocity-dependent. As spasticity is much more prominent in the lower limbs and its presence was found minimal in the distal arm, the influence of spasticity should not be overestimated for this study.[89, 90]

A considerable low reliability and validity was found for the choreoathetosis subscale in this protocol. The DIS was the first scale to set a standard for the clinical evaluation of this motor disorder and this study was the first to assess the concurrent validity of the DIS choreoathetosis subscale. More research is needed to ameliorate the current results for reliability and validity, which should lead to the ability to generalize the outcome of the SVM for choreoathetosis. In contrast, as good results were obtained in the dystonia subscale, it could be worth to explore the dystonia assessment tool as a single value.

This is a pioneer study using SVM to discriminate DYS and CA in a manually able DCP population, and is still in an explorative phase. Thus, more research is needed to confirm the obtained results. For this study, the SVM was trained in classifying a small number of classes with a large variation in the presentation of severity. To enable inclusion of severity variation, further classification and evaluation analyses can be pursued respectively using more refined classes and continuous data. This is possible using the relevant features obtained in this study as input in the SVM classifier and linear regression model.

5. Conclusion

This study was the first to validate automated dystonia and choreoathetosis discrimination during functional reaching tasks, showing promising outcomes.

The first part of this study analyzed the psychometric value of the DIS used in the IDCA protocol. The DIS was found a reliable and valid instrument to evaluate dystonia in the functional reaching tasks, with results being in line with previous research. However, poor results were found for the choreoathetosis subscale.

The second part of this study focused on the ultimate aim regarding the discriminative performance of the established SVM classifier model based on 3D objective motion data. Satisfying results were obtained for the combined classification of DYS and CA in a multiclass model, with a low prediction error for the DYS/No CA class. In a subsequent stage, highly accurate predictions were obtained in the classification of DYS and CA separately. A list of the unique discriminative characteristics for both motor disorders eventually could be made. Gyroscope data was prominent in differentiating and characterizing both DYS and CA. Accelerometer data was more related to dystonia whereas MOCAP features were more of interest in choreoathetosis. Time domain measures were principal in the discrimination of DYS and CA. Dystonia was more reflected by time domain measures, whereas frequency domain measures were of higher interest in the detection of choreoathetosis.

As results of the concurrent validity and interrater reliability study were not as good as expected for the choreoathetosis subscale, more research regarding the clinical evaluation of choreoathetosis is needed to ensure generalization of the obtained SVM results. In contrast, further investigation on a dystonia assessment tool is indicated, using the obtained features and more refined classes in the SVM classifier, and further extending towards a linear regression model with continuous data.

Further insights obtained in this study can assist correct treatment management plans, thereby improving quality of life in patients with DCP. Future research on automatic discrimination and evaluation of DYS and CA will allow opportunities to describe the motor disorders based on their biomechanical characteristics.

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Appendices

A. Demographic characteristics and DIS statistical analyses

Table A1: *Demographic characteristics of the participants*

Participant	Age	MACS	L/R handed	Measured side
P1	12.22y	3	R	L
P2	23.91y	1	R	L
P2	23.91y	1	R	R
P3	18.46y	2	L	L
P3 *	18.46y	3	L	R
P4	25.37y	3	L	R
P5	16.20y	2	R	L
P5	16.20y	2	R	R
P6	13.54y	2	L	R
P7	19.41y	2	R	L
P8	16.91y	3	R	L
P8	16.91y	3	R	R
P9	11.93y	2	R	L
P9	11.93y	2	R	R
P10	13.63y	3	R	L
P11	21.55y	3	R	L
P12	8.63y	2	L	R
	Mean Age 17.01y (SD 4.78y)			

* Eventually excluded from this study because of missing data

Table A2: *Interrater reliability of the DIS classifications (0-3): Intraclass Correlation Coefficients (ICC) with 95% confidence intervals (CI) between raters (k = 2) for the Dyskinesia Impairment Scale scored in functional reaching tasks.*

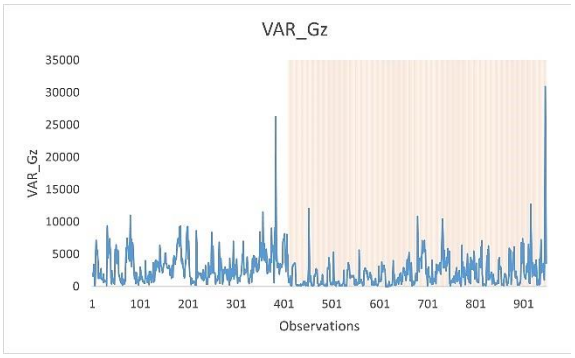
	ICC [95% CI]
RF1	0.172 [0.003-0.335]
RF2	0.356 [0.187-0.504]
RF3	0.344 [0.158-0.504]
RGV1	0.625 [0.490-0.729]
RGV2	0.236 [0.062-0.397]
RGV3	0.279 [0.073-0.459]
RS1	0.230 [0.060-0.389]
RS2	0.204 [0.035-0.364]
RS3	0.140 [-0.037-0.315]

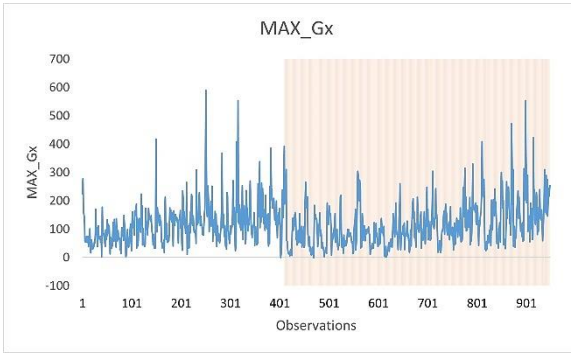
B. Machine learning classifier model

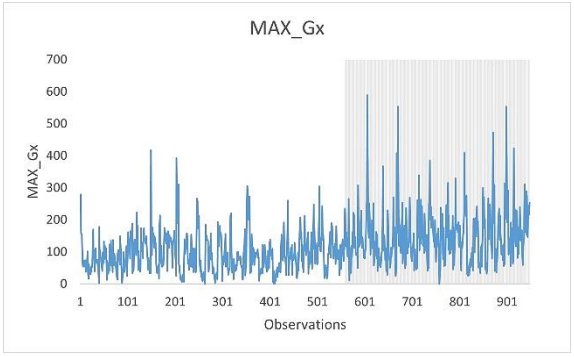
Table B1*: Relevant features per dataset*

MULTICLASS CLASSIFICATION	DYS CLASSIFICATION	CA CLASSIFICATION
MAX_Gx	VAR_Gz	MAX_Gx
MAX_Gy	MAX_Gx	MAX_GN
MAX_Gz	MAX_Gy	RMS_jerk_Gz
RMS_jerk_Gx	MAX_Gz	RMS_jerk_GN
RMS_jerk_Gy	RMS_jerk_Gx	Corr_Gxz
RMS_jerk_Gz	RMS_jerk_Gy	Max_vel_Ax
RMS_jerk_GN	RMS_jerk_GN	Max_vel_Ay
Cor_Gxy	STD_jerk_Az	Max_vel_Az
Cor_Gyz	Corr_Gxy	SM_AN
Cor_Gxz	Corr_Gyz	SM_Gx
Max_vel_Az	Max_vel_Az	SM_Gy
SM_Gx	SM_Ax	SM_Gz
SM_Gz	SM_Az	SM_GN
SM_Gy	SM_Gz	SpE_Gx
SM_Gz	SM_GN	SpE_Gy
SM_GN	SpE_Gy	FbandP2_Gx
SpE_Gy	SpE_GN	FbandP2_Gy
SpE_Gz	FbandP1_Ay	FbandP2_GN
SpE_GN	FbandP2_Gx	TrDev
FbandP2_Gx	FbandP4_Az	WrDev_min
FbandP2_Gy	TrDev	WrDev_max
FbandP2_Gz	WrFl_min	
TrDev		

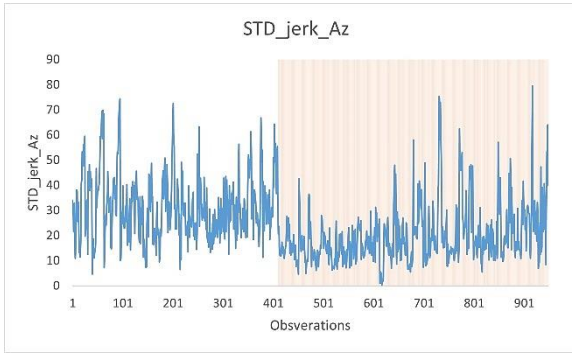
- Ct.1

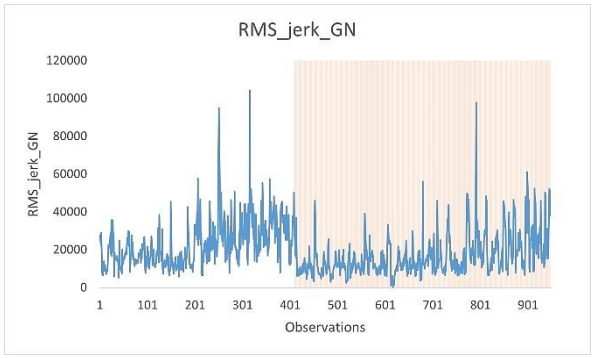






- Ct. 2





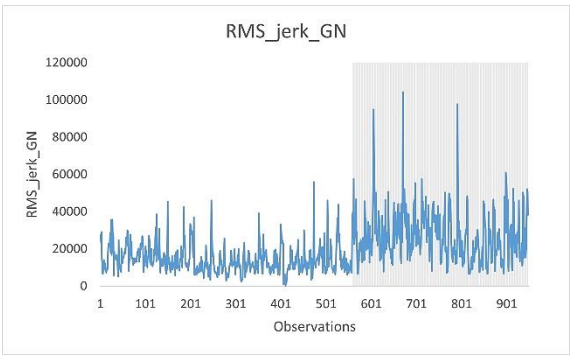
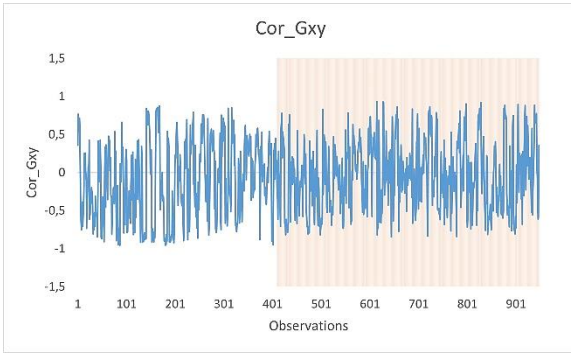
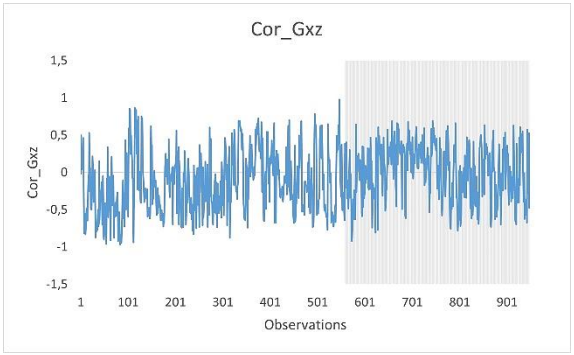


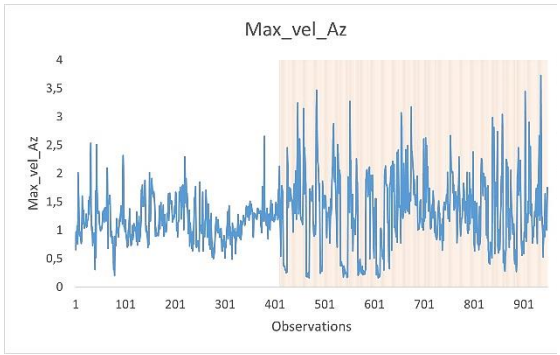
Figure B1: *Relevant feature plots of DYS and CA binary models*

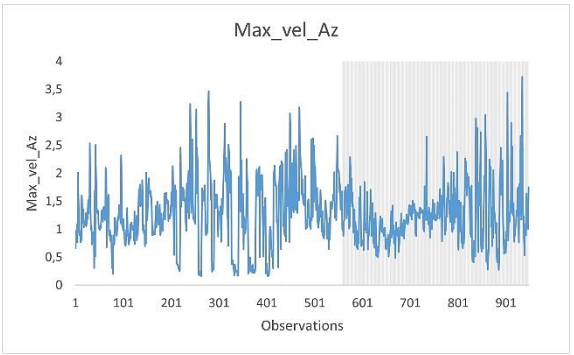
White = class 0 (No DYS or No CA); light orange marking = class 1 (DYS); light grey marking = class 1 (CA)

- Ct. 3

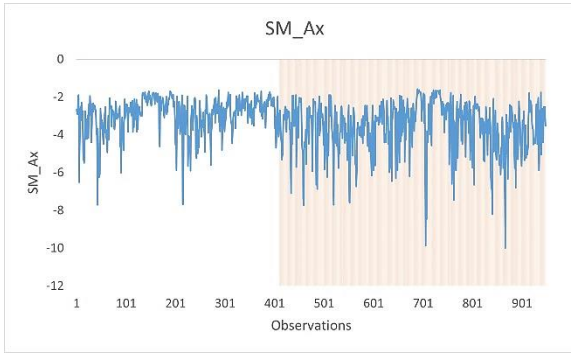


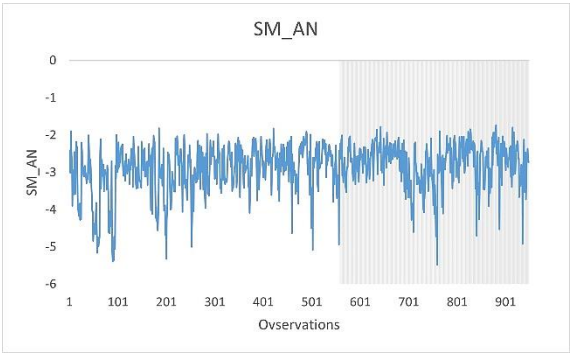


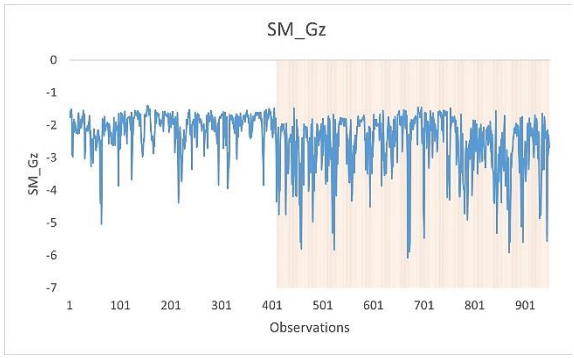




- Ct. 4







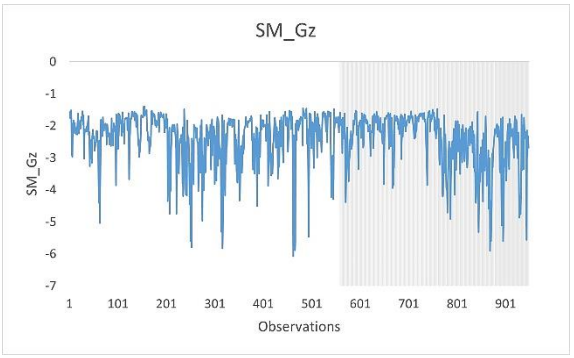
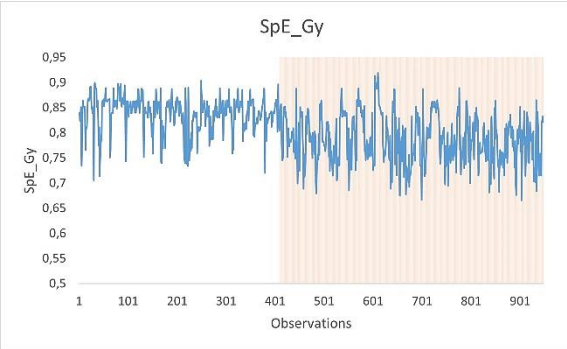
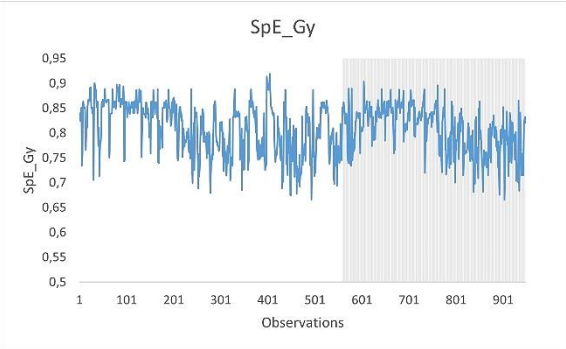


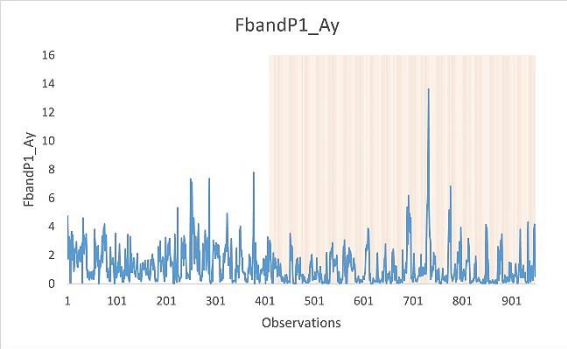
Figure B1: *Relevant feature plots of DYS and CA binary models*

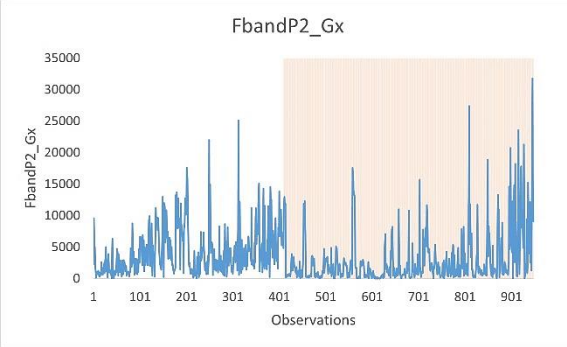
White = class 0 (No DYS or No CA); light orange marking = class 1 (DYS); light grey marking = class 1 (CA)

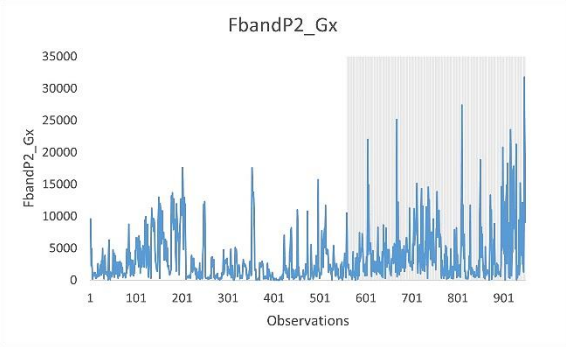




- Ct. 5







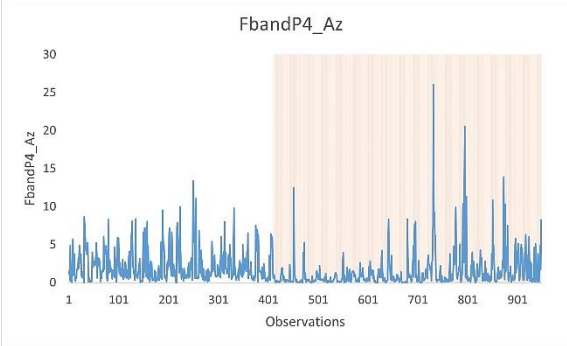
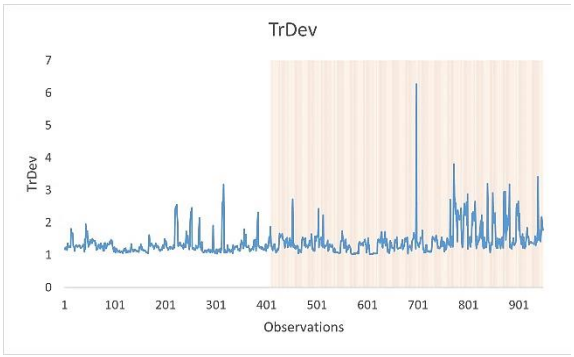
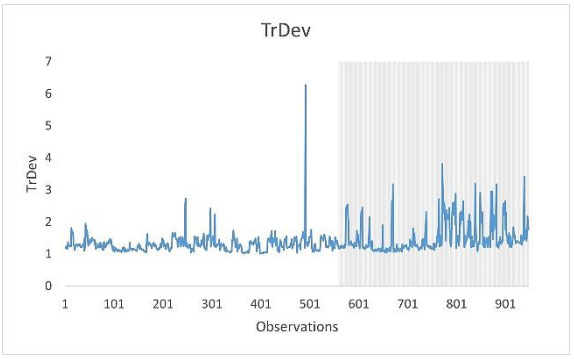


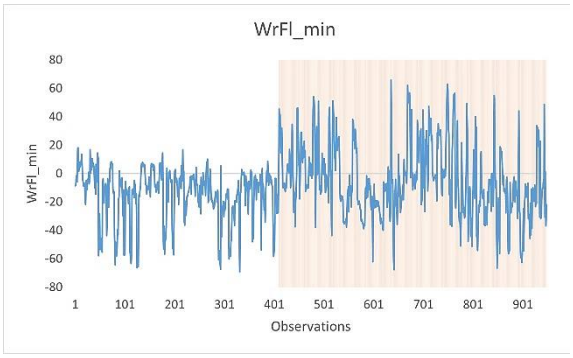
Figure B1: *Relevant feature plots of DYS and CA binary models*

White = class 0 (No DYS or No CA); light orange marking = class 1 (DYS); light grey marking = class 1 (CA)

- Ct. 6







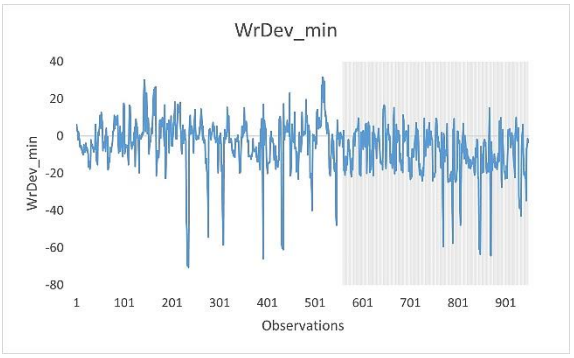
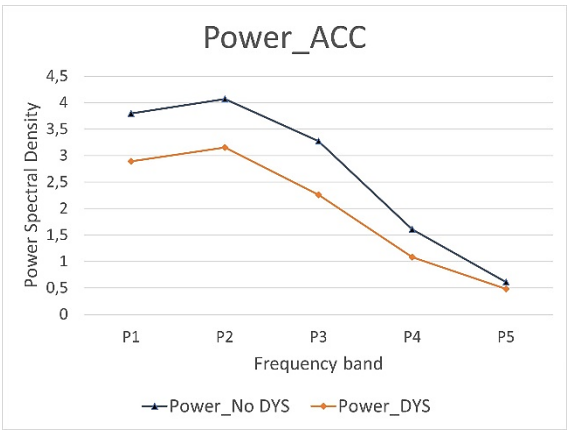
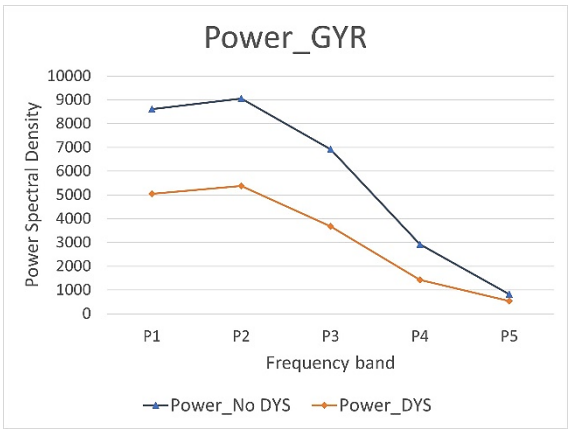


Figure B1: *Relevant feature plots of DYS and CA binary models*

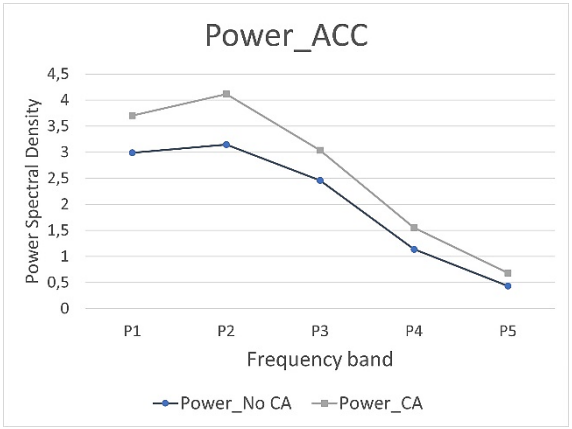
White = class 0 (No DYS or No CA); light orange marking = class 1 (DYS); light grey marking = class 1 (CA)

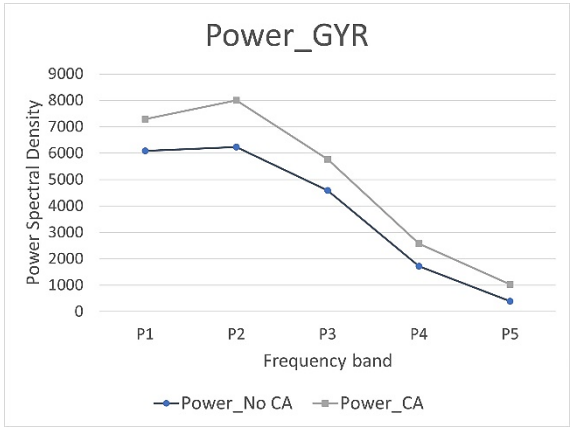
- No DYS / DYS



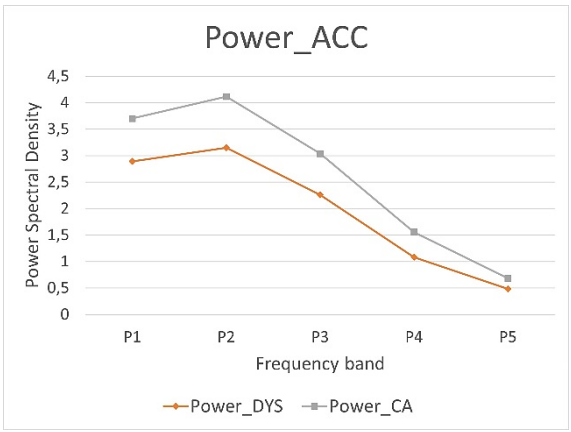


- No CA / CA





- DYS vs. CA



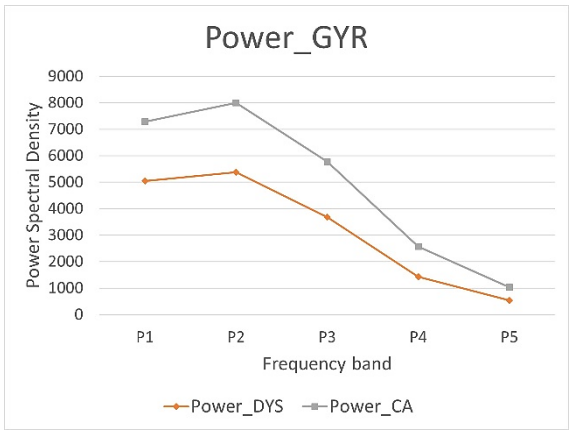


Figure B2: *Power spectrum of the DYS and CA binary classifier models and the comparison of DYS and CA*

C. Populaire samenvatting

Dyskinetische cerebrale parese (DCP) is een zeldzame motorische aandoening met schade aan het ontwikkelende brein. DCP uit zich klinisch in onwillekeurige bewegingen en moeilijk gecontroleerde vrijwillige bewegingen, ten gevolge van abnormale en wisselende spierspanningen.

Dystonie (DYS) en choreoathetose (CA) zijn twee omschreven bewegingsstoornissen binnen de DCP populatie. DYS wordt gekenmerkt door abnormale gewrichtstanden en CA door veelvoudig wringende bewegingen. Differentiatie en ernstbeoordeling van beiden zijn veelbetekenend in het doeltreffend toekennen van correcte behandeling. Het kan ook inzichten scheppen in het kennisluik rond de beperkt gekende aandoening, zijn therapie en prognose.

Differentiatie en evaluatie van DYS en CA wordt momenteel klinisch mogelijk gemaakt met de Dyskinesia Impairment Scale (DIS) als gouden standaard. Om reden van het deels subjectief en tijdrovende aspect van deze schaal en de verwachte ervaring, richtte onderzoek zich recent op objectieve meetinstrumenten. Deze zijn in staat gedetailleerd en betrouwbaar bewegingskarakteristieken weer te geven kenmerkend voor een bepaalde bewegingsstoornis. Binnen deze studie werd specifiek nagegaan of 3D bewegingskarakteristieken van het bovenste lidmaat gebruikt kunnen worden om dystonie en choreoathetose automatisch te gaan differentiëren.

Twaalf kinderen met DCP namen deel in een protocol bestaande uit drie taken: voorwaarts en zijwaarts reiken, en verticaal reiken en grijpen. De DIS werd gebruikt om DYS- en CA-aanwezigheid te scoren in de distale arm. Tijdens deze reiktaken werden de bewegingen vastgelegd door de registratie van polskinematica, voorarmversnelling en -hoeksnelheid middels markers en sensoren. De bekomen 144 bewegingsparameters werden toegevoegd in een support vector machine (SVM) learning classificatiemodel samen met hun klinische DIS classificaties. Deze studie vergeleek de voorspelde klassen van het SVM model met de vooraf bepaalde DIS classificaties. Hieropvolgend vond een selectieprocedure plaats om de relevante bewegingsparameters te identificeren voor beide bewegingsstoornissen apart. Als referentie voor het trainen en aftoetsen van de computergestuurde classificaties was een adequate schaal nodig. Hierom werd aanvullend de validiteit en betrouwbaarheid nagegaan van de DIS die in casu gebruikt werd bij het scoren van de functionele reiktaken.

Goede resultaten werden bekomen voor de DIS dystonie subschaal, namelijk matige tot excellente validiteit en matig tot goede betrouwbaarheid. Mindere resultaten werden verkregen voor choreoathetose; een afwezige tot matige validiteit en zwakke betrouwbaarheid. Resultaten wezen uit dat het SVM model met een redelijke nauwkeurigheid (74.58%) predictief onderscheid kan maken

tussen aan- of afwezigheid van DYS en CA. Betere resultaten werden gevonden in een SVM model dat enkel binair gaat differentiëren. Identificatie van de aan- of afwezigheid van DYS of CA is respectievelijk mogelijk met een 90.34% en 84.66% accuraatheid. Daarnaast werden typerende parameters opgelijst voor DYS en CA.

Deze studie was pionier in het valideren van een automatische DYS en CA discriminatie tijdens functionele taken van het bovenste lidmaat. De veelbelovende resultaten kunnen betekenisvol zijn in het creëren van nieuwe inzichten en het verbeteren van doelgerichte therapie. Komend onderzoek kan de klinische evaluatie van choreoathetose optimaliseren en de objectieve evaluatie van dystonie reeds verder uitbouwen.

D. Ethical approval

Bewegingspatronen bij kinderen en jongeren met dyskinetische cerebrale parese: een comprehensieve aanpak

Communicatiegeschiedenis

De verstrekte informatie toont aan dat het onderzoek in het kader van de masterproef integraal deel uitmaakt van een studie die reeds werd goedgekeurd door een erkend ethisch comité.

U hoeft bijgevolg geen nieuw dossier in te dienen in het kader van uw masterproef. Wel dient u het UZ/KU Leuven toe te voegen als nieuw deelnemend centrum aan het huidige dossier indien dit nog niet het geval was. Gelieve u bij het betreffende comité te informeren en u te conformeren aan de daar geldende procedures en hun adviezen te volgen.

Subject to the completeness and accuracy of the details you provided, take this as confirmation that you may begin the study as part of the Master's thesis.

We would like to point out that you remain responsible for your own research. Furthermore any change in the information you provided about the research design renders any previous approval null and void. If this is the case, you must amend your current file and present it to the Committee that previously approved it.

Details met de antwoorden van de vragenlijst zijn hier raadpleegbaar.

Download resultaat ethische toetsing

Dossiernr.	MP013300
Stage **Faculteit/opleiding**	Masterproef FaBeR Faculteit Bewegings- en Revalidatiewetenschappen - LOBW-REVAKI-REHAB-IMAPA-SGK-EMA
Studiefase **Academiejaar**	eerste fase 2019-2020
Korte omschrijving / abstract	Introductie: Dyskinetische cerebrale parese (DCP) is de tweede meest voorkomende groep binnen CP. Dystonie en choreo-athetose zijn de 2 bewegingsstoornissen die dominant aanwezig zijn bij DCP. Deze bewegingsstoornissen zijn complex met een grote negatieve impact op het dagelijks functioneren. Ondanks het ernstig limiterende effect van dystonie en choreo-athetose in het dagelijkse leven, staat de behandeling van deze complexe bewegingsstoornissen nog in de kinderschoenen. Therapeutische behandeling is op dit moment vooral gebaseerd op klinische ervaring, zonder wetenschappelijk onderbouwde achtergrond. Medische behandeling is moeilijk, omdat sommige medicatie werkt voor dystonie en niet voor choreo-athetose en vice-versa. Het onderscheid tussen dystonie en choreo-athetose wordt echter te weinig gemaakt, ondanks een cruciale factor zijnde in het de keuze van behandelingsstrategie. Drie-dimensionele bewegingsanalyse kan een mogelijkheid bieden om deze complexe bewegingsstrategieën op een objectieve manier te analyseren. Dit is een vaak gebruikte techniek voor het opvolgen van bewegingen en van daaruit gewrichtshoeken en hoeksnelheden te berekenen. Dit protocol is echter gebonden aan specifieke daarvoor ontwikkelde camera's, wat de gebruiksvriendelijkheid en,van niet ten goede komt. Sensoren hebben de laatste jaren meer aandacht gekregen in de wereld van bewegingsanalyse omdat deze devices klein, mobiel en gebruiksvriendelijk zijn. Sensoren meten lineaire versnelling en hoeksnelheid tijdens een beweging. Deze devices kunnen eveneens gebruikt worden om bewegingen te registreren en zijn niet locatie-gebonden. De rechtstreekse registratie van versnelling biedt mogelijkheden bij een populatie waarbij de coördinatie sterk verstoord is. Het doel van dit onderzoek is om dystonie en choreo-athetose te discrimineren bij kinderen en jongeren met dyskinetische CP op basis van drie-dimensionele data tijdens bewegingen van het bovenste lidmaat. Dit onderzoek draagt bij tot het optimaliseren van de behandelingsstrategieën van complexe bewegingsstoornissen in kinderen en jongeren met dyskinetische CP. Hypothese: De hypothese is dat dystonie en choreo-athetose onderscheiden kunnen worden op basis van hun respectievelijke biomechanische parameters. Methodologie: De data verzameld in dit onderzoek bestaat uit de biomechanische bewegingsparameters van sensoren en infra-rood camera's, en videobeelden van de Dyskinesia Impairment Scale, een video-gebaseerde meetschaal. De verzamelde biomechanische parameters zullen gebruikt worden om via geavanceerde data analyses dystonie en choreo-athetose te onderscheiden. De dystonie en choreo-athetose % scores van de Dyskinesia Impairment Scale zullen als validatie voor deze discriminatie gebruikt worden. Praktisch: Datacollectie gebeurt in de KU Leuven campus Brugge en campus Pellenberg.
Opmerkingen / praktische informatie	**Taken:** - Tijdlijn maken (november 2019 - juni 2021) - 1st year seminar: mei 2020 (literatuurstudie + methodology): powerpoint presentatie - Literatuurstudie - Assistentie tijdens labo testen - Data analyse - Statistische analyse Statistische analyse zal gebeuren in SPSS. - Schrijfproces
Website	https://gbiomed.kuleuven.be/english/research/50000743/nrrg1/nrrg.htm#Elegast%20Projects
Toegepaste technieken	3D bewegingsanalyse ; sensor data analyse
Key publication	Clinical presentation and management of dyskinetic cerebral palsy Elegast Monbaliu, Kate Himmelmann, Jean-Pierre Lin, Els Ortibus, Laura Bonouvrié, Hilde Feys, R Jeroen Vermeulen, Bernard Dan The Lancet Neurology
Trefwoorden	cerebral palsy · dyskinetic cerebral palsy · movement analysis · biomechanics · Movement disorders
Gewenste taal van communicatie	nl
Promotor **Copromotor** **Begeleider/Contact**	Professor Elegast Monbaliu (u0056375) Mejuffrouw Inti Vanmechelen (u0102306) Dr Marco Konings (u0126419) Mejuffrouw Inti Vanmechelen (u0102306)
Organisatie: **Dienst:** **Discipline:**	KU Leuven Revalidatiewetenschappen, Campus Brugge Masterproef
Extra groepsleden	 Ellen Van Wonterghem ellen.vanwonterghem@student.kuleuven.be r0651230  Sophie Gardyn sophie.gardyn@student.kuleuven.be r0632953

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Richtlijnen voor auteurs voor publicaties van de ‘guidelines for authors’ in Gait and Posture:

https://www.elsevier.com/journals/gait-and-posture/0966-6362/guide-for-authors