Annals of Physical and Rehabilitation Medicine 62 (2019) 409–417

Available online at ScienceDirect www.sciencedirect.com

Elsevier Masson France EM|consulte www.em-consulte.com

Original article

Detection of pronator muscle overactivity in children with unilateral spastic cerebral palsy: Development of a semi-automatic method using EMG data

Aurélie Sarcher [a,b,*], Sylvain Brochard [b,c], Brigitte Perrouin-Verbe [a], Maxime Raison [d], Guy Letellier [e], Fabien Leboeuf [a,f], Raphaël Gross [a,g]

[a] Motion analysis laboratory, physical medicine and rehabilitation, university hospital of Nantes, 44000 Nantes, France
[b] Laboratory of medical information processing (LaTIM), INSERM UMR 1101, 29200 Brest, France
[c] Physical medicine and rehabilitation, university hospital of Brest, 29200 Brest, France
[d] Rehabilitation engineering chair applied to pediatrics (RECAP), Sainte-Justine university hospital and école polytechnique de Montréal, Montréal, QC, Canada
[e] Pediatric rehabilitation center ESEAN, 44200 Nantes, France
[f] College of health and social care, university of Salford, Salford, United Kingdom
[g] Laboratory Movement-Interactions-Performance (MIP), EA 4334, university of Nantes, 44322 Nantes, France

ARTICLE INFO

Article history:
Received 7 September 2018
Accepted 1st August 2019

Keywords:
Cerebral palsy
Children
Electromyography
Forearm supination
Overactivity
Pronators

ABSTRACT

Background: The pronator teres and pronator quadratus muscles are frequently injected with neuromuscular blocking agents to improve supination in children with spastic cerebral palsy and limited active elbow supination. However, determining by simple clinical examination whether these muscles are overactive during active movement is difficult.
Objective: This study aimed to develop a semi-automatic method to detect pronator muscle overactivity by using surface electromyography (EMG) during active supination movements in children with cerebral palsy.
Methods: In total, 25 children with unilateral spastic cerebral palsy (10 males; mean [SD] age 10.6 [3.0] years) and 12 typically developing children (7 males; mean age 11.0 [3.0] years) performed pronation–supination movements at 0.50 Hz. Kinematic parameters and surface EMG signals were recorded for both pronator muscles. Three experts visually assessed muscle overactivity in the EMG signals of the children with cerebral palsy, in comparison with the reference group. The reliability and discrimination ability of the visual assessments were analysed. Overactivity detection thresholds for the semi-automatic method were adjusted by using the visual assessment by the EMG experts. The positive and negative predictive values of the semi-automatic detection method were calculated.
Results: Intra-rater reliability of visual assessment by EMG experts was excellent and inter-rater reliability was moderate. For the 25 children with unilateral spastic cerebral palsy, EMG experts could discriminate different profiles of pronator overactivity during active supination: no pronator overactivity, one overactive pronator, or overactivity of both pronators. The positive and negative predictive values were 96% and 91%, respectively, for this semi-automatic detection method.
Conclusions: Detection of pronator overactivity by using surface EMG provides an important complement to the clinical examination. This method can be used clinically, with the condition that clinicians be aware of surface EMG limitations. We believe use of this method can increase the accuracy of treatment for muscle overactivity, resulting in improved motor function and no worsening of paresis.

1. Introduction

Unilateral spastic cerebral palsy (USCP) is characterised by spastic paresis that impairs motor control of both upper and lower limbs on one side of the body [1]. Gracies (2005) classified the motor symptoms related to spastic paresis [2], and suggested that

* Corresponding author at: Physical medicine and rehabilitation, motion analysis laboratory, university Hospital of Nantes, 44093 Nantes, France.
E-mail address: aurelie.sarcher@gmail.com (A. Sarcher).

https://doi.org/10.1016/j.rehab.2019.08.001
1877-0657/

3 symptoms particularly limit active movement in children with USCP:

- paresis of the agonist muscles (i.e., lack of recruitment of the agonist muscles during active movement);
- contracture of the antagonist muscles (i.e., permanent shortening of the muscles, which restricts the range of motion [ROM]);
- and overactivity of the antagonist muscles during active movement, resulting in spastic coactivation [2–4]. Not all children with USCP have all of these symptoms, and their presence and severity vary depending on the movement being performed.

Clinically, diagnosing muscle overactivity during active movement is difficult. Clinical evaluations commonly include a rating of spasticity with the Modified Ashworth Scale (MAS) [5] or the Modified Tardieu Scale [6]. However, spasticity is defined as a short excessive response of a muscle to a passive stretch and may have no direct relation to muscle responses that occur during active movement [2]. Moreover, it is not possible to determine whether all muscles within a functional group are overactive or if just one is overactive.

Despite these issues, movement impairments are commonly treated by injecting neuromuscular blocking agents into the antagonist muscles [1,2,7] in an attempt to improve active movement. However, this treatment may actually increase paresis of the injected muscles [8]. Therefore, it should be performed only in muscles in which overactivity during active movement has been confirmed and should not solely be based on spasticity found during the clinical examination.

One objective method to determine the presence of overactivity during active movements is electromyography (EMG). However, the interpretation of EMG measurements can be challenging because of high levels of signal variability. Interpretation is best carried out by "expert" clinicians who are experienced in EMG. EMG is more commonly used in clinical research, and variables that are frequently extracted from the EMG signals include duration, amplitude and timing of activation. EMG has been used in both research and clinical practice to evaluate these variables in the lower-limb muscles of children with USCP [3,9–11] but has seldom been used in the upper limbs [12–15] even though active upper-limb movement is frequently limited in children with USCP [1,16]. In particular, children with USCP often have difficulty performing forearm supination [15,17]. The movement of supination is regulated by 2 agonist muscles (biceps brachii and supinator) and 2 antagonist muscles (pronator teres and pronator quadratus). One or both of the pronator muscles being excessively active during supination can limit supination.

One study [13] found excessive activation of both pronator teres and pronator quadratus muscles during active supination in children with USCP as compared with typically developing (TD) children [13]. Moreover, the study found an association between excessive coactivation of pronator/supinator muscle pairs during supination and limited active range of supination. This work confirmed that pronator muscle overactivity during supination can reduce the range of active supination. The next step was to verify whether EMG experts could:

- reliably analyse the EMG patterns of both pronator muscles, as is usual practice in the clinic;
- differentiate between various pronator-muscle overactivity profiles in children with USCP.

Recently, Sarcher et al. [18] developed an individualized method to detect differences between surface EMG signals from a method initially developed by Schwartz et al. [19] and modified by Chia and Sangeux [20]. The method displays the EMG patterns, with their variability, directly on screen, and an integrated statistical tool highlights significant differences between patterns. We believe this method could help inexperienced clinicians interpret EMG signals, but it has not yet been validated by comparison with a gold-standard method such as the interpretation of EMG signals by experts.

The overall aim of this study was to develop a surface EMG semi-automatic method of detecting pronator muscle overactivity in children with spastic CP by using data from visual EMG assessment by experts. The primary objective was to establish the inter-rater and intra-rater reliability of visual assessment by EMG experts of pronator muscle overactivity in surface EMG signals. The secondary objective was to establish the ability of EMG experts to discriminate between pronator muscle overactivity profiles during active supination movements in children with USCP by using surface EMG, based on the hypothesis of the existence of different profiles. The third objective was to adjust the overactivity detection thresholds of the semi-automatic method by using the results of the visual assessments by the EMG experts, considered the gold standard, to ensure a high concurrent validity of the method.

2. Methods

2.1. Participants

We included a convenience sample of 32 children with USCP (16 males; mean [SD] age 10.7 [3.0] years, range 6.2–17.4) who had different degrees of upper-limb disability. We excluded children with total active range of supination (from maximal pronation to maximal supination) $< 20^\circ$ because the method of EMG detection presented in this paper required a minimum range of active movement for identifying the phases of pronation and supination. The other exclusion criteria were restriction of passive supination $> 10^\circ$ (indicating contracture of one or both pronator muscles); botulinum toxin injections within the previous 6 months; previous upper-limb surgery to the affected upper limb, and inability to understand or perform the experimental tasks. Of the 32 children with USCP originally identified, 7 were excluded because they had a total active range of supination $< 20^\circ$ on motion analysis (kinematic data). Data for 25 children were analysed.

The Manual Ability Classification System [21] was used to grade manual ability [1,quite good; 5, very impaired]. All 25 children with USCP had a Manual Ability Classification System level of 1 ($n = 11$) or 2 ($n = 14$). The Modified Ashworth Scale (MAS) [5] was used to rate pronator muscle spasticity (0, none; 4, severe). MAS scores for the 25 children ranged from 0 to 3.

The reference group consisted of 12 TD children.

Our local ethics committee Comité de Protection des Personnes Ouest IV (France) approved the study (IdRCB no. 2016-A01314-47). Written informed consent was obtained from both parents of each child, and informed assent was obtained from all children. Data were processed in accordance with the requirements of the French National Commission for Data Protection and Liberties (CNIL).

2.2. Experimental set-up

In the USCP group, only data for the affected sides were analysed. In the reference group (TD children), the side to be analysed was randomly chosen (dominant side analysed for $n = 4$ and non-dominant side for $n = 8$).

2.2.1. Kinematic parameters

Participants were equipped with 29 reflective markers on their hand, forearm, upper arm, shoulder and thorax, according to the upper-limb model developed by Laitenberger et al. [22]. Displacement of the markers was recorded by using a 3-D motion capture system (VICON, Oxford Metrics, Oxford, UK) at a sampling frequency of 100 Hz.

2.2.2. EMG

Activity of pronator teres and pronator quadratus muscles was recorded by using self-adhesive pairs of disposable bipolar Ag/AgCl surface EMG electrodes (recording diameter 10 mm). EMG signals were recorded with 2 different wireless surface EMG systems because of a change of equipment during the study: a FreeEMG300 system (BTS, Milan, Italy) used for 9 USCP participants (system 1) and a Cometa ZeroWire system (Cometa, Milan, Italy) for 16 USCP and 12 TD participants (system 2). In all cases, the sampling frequency was 1000 Hz and the systems were synchronized with the motion capture system.

Fig. 1 shows the placement of the electrodes over pronator teres and pronator quadratus muscles. To determine the placement of the electrodes, the pronator teres muscle was palpated during isometric voluntary contractions against manual resistance of forearm pronation [23,24], and the surface EMG electrode was placed on the most prominent part of the muscle. For the pronator quadratus, the surface EMG electrode was placed on the anterior aspect of the forearm, perpendicular to the forearm axis, about one finger proximal to the wrist crease [23,25].

2.3. Procedure

Participants were seated on a height-adjustable bench. The baseline EMG signal was recorded for several seconds while the child sat relaxed (at rest). An EMG expert confirmed the stability of the baseline signal before the child was asked to perform at least 5 consecutive movement cycles: extension–flexion (EF) in the sagittal plane (without shoulder elevation) and forearm pronation–supination (PS). The children were asked to perform active movements to their maximal active ROM (AROM) and to keep the other joints as still as possible during each trial. The movement velocity was regulated by an auditory metronome at a fixed frequency of 0.50 Hz to ensure similar conditions between children because muscle activity during active movement increases with movement velocity in children with and without USCP [10,12].

2.4. Data processing

Data processing involved using custom MATLAB routines (Mathworks, Natick, MA, USA) and the open-source Biomechanical ToolKit library [26].

2.4.1. Kinematic parameters

Joint kinematics were obtained by using a refined subject-specific model of the upper limb [22] and an inverse kinematic process combined with a global optimization [27]. Maximal supination angle (°) and AROM from maximal pronation to maximal supination (°) were extracted from the PS angles. To evaluate a risk of crosstalk between forearm muscles, the ROM of elbow EF, wrist EF, and wrist ulnar/radial deviation (°) during active supination were also extracted.

2.4.2. EMG processing

EMG signals were band-pass-filtered (10–450 Hz, Butterworth zero-lag 4th order), full-wave–rectified and smoothed with a low-pass filter (50 Hz, Butterworth zero-lag 2nd order). An activation threshold was used as recommended [28] and according to previously published protocols [12,13]. EMG signals were linearly interpolated to 100 points to represent the full movement cycle. For PS, the first half of the movement was pronation, and the second half was supination.

EMG signals were amplitude-normalized with the peak value measured over the averaged movement trials (i.e., the averaged EF trial and the averaged PS trial) as recommended by Sarcher et al. [18]. Indeed, Sarcher et al. [18] found that this normalization, when used in a similar population and for similar movements as those in the present paper, resulted in higher inter-session reliability than normalization by the peak value during maximal voluntary contractions or the peak value measured over all movement trials. Moreover, the risk of overestimating the muscle activation of a paretic muscle might be amplified when using a normalization method involving strong contractions in children with CP, because they are unable to adequately activate their muscles voluntarily.

2.5. Calculation of measurement variability

The variability of the kinematic and EMG measurements was calculated as described by Schwartz et al. [19] and modified by Chia and Sangeux [20]. This method was already applied to upper-limb data by Sarcher et al. [18] and consists of calculating the variability of the data:

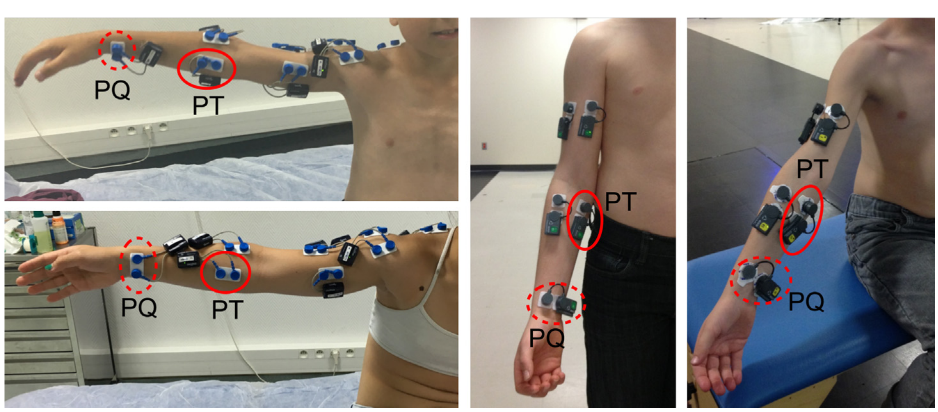

Fig. 1. Examples of 4 children equipped with surface electromyographic electrodes placed over the pronator teres (PT) and pronator quadratus (PQ) muscles. On the left, 2 children are equipped with system 2, a Cometa ZeroWire system (Cometa, Milan, Italy), and on the right, 2 children are equipped with system 1, a FreeEMG300 system (BTS, Milan, Italy).

- for the averaged signal of the reference (TD) group, including inter-trial and inter-session variability (EMG signals were recorded for the TD children during 2 different sessions) and inter-subject variability;
- for each child with USCP, including the child's own inter-trial variability and the inter-session variability of a group of children with USCP [18].

Variability was calculated for each time point of the movement cycle, allowing for direct visual comparison of the EMG signal for each child with USCP with that for the reference group, for each time point of the movement cycle.

2.6. Visual assessment of pronator muscle overactivity by EMG experts

A group of 3 EMG experts (1 physical and rehabilitation medicine physician and 2 biomechanical engineers with several years' experience in movement analysis, including dynamic EMG recordings) visually assessed both the rectified signals and the post-processed signals for the 50 pronator muscles of the 25 children with USCP during the PS cycle and compared signals with those of the reference group. EMG experts were asked to classify each signal as "normal" if they determined that the pronator muscle activation during supination was similar to that of the reference group, "overactive" if they determined that the pronator muscle activation during supination was clearly higher than that of the reference group, "borderline" if they determined that the pronator muscle activation during supination had a tendency to be higher than that of the reference group but the tendency was not constant, or "undefined" if they found that the pronator muscle activation during supination could not be adequately interpreted. The assessments were performed independently by each EMG expert, and they had no access to any other data from the children during the assessments. The EMG experts rated the signals on 2 occasions, at least 1 week apart, for analysis of intra-rater reliability.

2.7. Surface EMG semi-automatic detection method

The surface-EMG semi-automatic detection method produced a display of the EMG patterns with their variability along with a statistical test that highlighted significant differences between EMG patterns, as developed by Schwartz et al. [19]. This test was applied to each time point of the movement cycle and included the calculation of data variability. Statistical significance was set at $P < 0.05$ [19]. The results indicated whether the EMG signal of the child with USCP was statistically higher, lower or within the range of the EMG profile of the reference group (TD children) for each time point of the movement cycle. However, this statistical tool is only indicative and there are currently no published data on the relation between the duration of statistically excessive activation and overactivity as defined by EMG experts. Therefore, the overactivity detection thresholds were adjusted by using the visual assessment by the EMG experts, considered the gold standard.

Statistical analysis

The inter- and intra-rater reliability of the EMG experts' assessment was evaluated by using percentage agreement (i.e., the proportion of assessments with agreement) and the Krippendorff's alpha reliability estimate with 95% confidence intervals (CIs) [29,30]. Krippendorff's alpha is an alternative to Cohen's kappa for data with multiple categories and multiple evaluators. It represents the ratio of observed to expected disagreement. Because 2 EMG recording systems were used in the study, we verified whether the system used (1 or 2) influenced the EMG expert evaluations by using the Fisher-Freeman-Halton extension of the Fisher exact probability test [31].

Concurrent validity of the surface-EMG semi-automatic detection method was assessed by using percentage agreement and the Krippendorff's alpha reliability estimate with 95% CIs. Positive and negative predictive values were also calculated.

The test statistic developed in Schwartz et al. [19] was also used to identify a significant difference between the kinematic data for each child with USCP and the TD group for ROM from maximal pronation to maximal supination (°), maximal supination angle (°), and ROM of elbow EF, wrist EF and wrist ulnar/radial deviation (°) during active supination. Statistical significance was set at $P < 0.05$.

3. Results

We included 25 children with USCP (10 males; mean [SD] age 10.6 [3.0] years, range 6.2–17.4) and 12 TD children (7 males; mean age 11.0 [3.0] years, range 6.7–15.9).

3.1. Kinematic parameters

The 25 children with USCP had a total active range of supination (from maximal pronation to maximal supination) of 21 to 148° (mean [SD] 73° [34°]) (Table 1). The range of elbow flexion during supination was significantly increased in 3 children with USCP as compared with the reference (TD) group and range of wrist radial deviation during supination was significantly increased in 3 children with USCP as compared with the TD group. The range of wrist flexion/extension during supination did not differ between any child with USCP and the TD group.

3.2. Intra- and inter-rater reliability of EMG expert evaluations

Krippendorff proposed that an alpha value ≥ 0.80 represents excellent agreement, and 0.67 is the lowest conceivable limit for considering agreement [30] (Table 2). In our study, intra-rater reliability was excellent, with percentage agreement 91%, and Krippendorff's alpha 0.85 (95% CI 0.77–0.93). Inter-rater reliability was moderate, with percentage agreement 82%, and Krippendorff's alpha 0.69 (0.50–0.87).

3.3. Discriminative ability of the EMG experts

The 3 EMG experts analysed 50 EMG pronator muscle signals ($n = 25$ pronator teres and $n = 25$ pronator quadratus) for children with USCP (Appendix 1). For 38/50 pronator signals, the 3 EMG experts showed complete agreement (comparison of the first evaluation by each assessor) (18 overactive; 20 normal). For 9/50 pronator signals, 2 of the 3 experts agreed (7 overactive; 1 normal; 1 undefined); therefore, the activity of these muscles was defined according to the assessment by these 2 experts. For 3/50 pronator signals, all 3 experts disagreed between borderline, normal and overactive; therefore the activity of those muscles was defined as borderline.

The conclusions of the EMG experts were that 25 pronator muscles were overactive (17 pronator teres; 8 pronator quadratus), 21 had normal activation (7 pronator teres; 14 pronator quadratus), 3 were borderline (1 pronator teres; 2 pronator quadratus), and 1 was undefined (1 pronator quadratus).

With systems 1 and 2, the ratio of overactive, normal, undefined and borderline pronator muscles was 11/18 (61%) and 14/32 (44%); 6/18 (33%) and 15/32 (47%); 0/18 (0%) and 1/32 (3%); and 1/18 (6%) and 2/32 (6%), respectively. The test statistic of the Fisher-Freeman-Halton extension of the Fisher exact probability test was

Table 1
Data for the 25 children with unilateral spastic cerebral palsy (USCP).

Children with USCP	Age (yr)	Sex	Side of USCP	MACS	MAS	Elbow PS AROM during supination	Supination max angle (?)	Pronator teres overactivity		Pronator quadratus overactivity		Elbow FE AROM during supination	Wrist FE AROM during supination	Wrist ulnar/ radial deviation AROM during supination	EMG system
					Pronators	Mean (SD)	Mean (SD)	EMG Experts	% excessive activation	EMG Experts	% of excessive activation	Mean (SD)	Mean (SD)	Mean (SD)	
1	10.6	F	L	1	1	100 (8)***	48 (8)**	0	0	0	0	8 (5)	19 (8)	5 (2)	2
2	11.0	F	L	1	1+	73 (9)***	30 (7)***	1	0	0	0	9 (5)	10 (6)	6 (3)	2
3	9.1	F	L	1	0	93 (7)***	22 (7)***	1	17	1	7	8 (5)	29 (6)	15 (2)	2
4	16.5	F	L	2	Missing	33 (6)***	−12 (7)***	1	5	1	26	6 (4)	7 (4)	7 (2)	2
5	10.9	M	R	2	Missing	26 (10)***	12 (9)***	1	18	Undefined	9	18 (6)	5 (4)	2 (2)	2
6	16.5	F	R	2	2	82 (6)***	19 (7)***	1	11	0	0	27 (7)** (Flexion)	7 (4)	15 (2)	2
7	17.4	M	R	1	0	115 (6)*	72 (7)	1	10	0	1	7 (5)	14 (5)	25 (3)* (Radial dev)	2
8	6.8	M	R	1	0	105 (7)***	51 (7)**	1	21	0	0	12 (5)	28 (8)	23 (3)	1
9	7.5	M	L	2	1+	50 (7)***	29 (7)***	1	28	1	14	7 (4)	11 (5)	26 (3)* (Radial dev)	1
10	9.1	F	R	1	1	83 (8)***	55 (8)*	1	3	0	0	10 (5)	25 (8)	7 (3)	1
11	9.2	M	R	2	1+	43 (7)***	25 (7)***	1	26	1	22	6 (5)	6 (4)	8 (3)	1
12	11.4	M	R	1	1	74 (7)***	46 (8)**	0	0	1	25	10 (5)	21 (9)	8 (4)	1
13	6.2	F	L	1	0	148 (6)	88 (7)	0	0	0	0	17 (6)	17 (6)	13 (4)	1
14	10.8	F	L	2	1	95 (11)***	69 (7)	Borderline	7	1	7	10 (5)	14 (8)	16 (2)	1
15	12.0	F	L	2	1	114 (8)*	69 (7)	0	0	1	15	10 (6)	22 (8)	15 (4)	1
16	7.7	F	L	2	1+	63 (8)***	1 (7)***	1	15	1	9	17 (6)	23 (6)	8 (2)	1
17	8.4	M	R	2	2	21 (7)***	−26 (7)***	1	36	Borderline	2	89 (7)*** (Flexion)	19 (7)	10 (3)	2
18	10.6	M	L	1	0	92 (8)***	51 (7)*	1	27	Borderline	1	17 (6)	22 (10)	12 (5)	2
19	8.6	F	R	1	0	83 (7)***	60 (7)*	0	0	0	0	3 (4)	28 (11)	30 (3)* (Radial dev)	2
20	8.0	M	R	1	0	122 (6)*	76 (8)	0	0	0	0	11 (5)	14 (6)	8 (4)	2
21	10.0	F	R	2	Missing	63 (7)***	56 (8)*	1	3	0	0	29 (6)*** (Flexion)	13 (7)	12 (4)	2
22	13.4	F	R	2	Missing	45 (7)***	2 (7)***	1	26	0	0	17 (5)	7 (4)	7 (2)	2
23	13.0	F	L	2	1+	40 (7)***	−5 (7)***	1	0	0	0	7 (4)	13 (6)	9 (2)	2
24	11.3	M	L	2	3	25 (6)***	−32 (7)***	1	31	0	0	8 (6)	8 (5)	3 (2)	2
25	7.9	F	R	2	1+	47 (7)***	20 (8)***	0	0	0	0	13 (5)	15 (5)	7 (3)	2
Reference group	11.0	–		–	–	141 (8)	79 (6)	–	–	–	–	8 (3)	15 (8)	14 (6)	2

The Manual Ability Classification System (MACS) was used to grade manual ability [1,quite good; 5, very impaired] (Eliasson et al., 2006) and the Modified Ashworth Scale (MAS) was used to rate the spasticity of the pronators [0, none; 4, severe] (Bohannon et al., 1987). The following variables were extracted from kinematic data: maximal supination angle (°) (0° = neutral PS; negative values = pronation; positive values = supination); ranges of motion of elbow flexion/extension (FE), wrist FE and wrist ulnar/radial deviation during supination (°). Pronator electromyography (EMG) data for the children with USCP during supination were compared to those of the reference group by EMG experts: each pronator was defined as "overactive" (1), "normal" (0), "borderline" or "undefined" based on EMG activation and decisions between EMG experts. Those data were also compared to those of the reference group by using the statistics described in both Schwartz et al. (2004) and the present article: the duration (%) when the EMG signal amplitude was significantly ($P < 0.05$) higher than that of the reference group during supination is indicated. AROM: active range of motion; F: female; M: male; MACS: Manual Ability Classification System; MAS: Modified Ashworth Scale; PS: pronation–supination. EMG system: a FreeEMG300 system (BTS, Milan, Italy) (system 1) and a Cometa ZeroWire system (Cometa, Milan, Italy) (system 2).

* $P < 0.05$.
** $P < 0.005$.
*** $P < 0.0005$.

Table 2
Intra- and inter-rater reliability for the detection of overactivity by EMG experts, evaluated by using the percentage of agreement (%) and Krippendorff's alpha reliability estimate with 95% confidence intervals (CIs).

Reliability	All EMG experts	EMG expert		
		A	B	C
Intra-rater				
Agreement (%)	91	96	92	86
Krippendorff's alpha (95% CI)	0.85 (0.77–0.93)	0.93 (0.84–1.00)	0.85 (0.71–0.99)	0.75 (0.58–0.92)
Inter-rater				
Agreement (%)	82			
Krippendorff's alpha (95% CI)	0.69 (0.50–0.87)			

$P = 0.815$. Therefore, the visual assessments by the EMG experts were not influenced by the EMG system used to record muscle activation.

3.4. Profiles of pronator muscle overactivity in children with USCP

The following profiles of pronator muscle overactivity were defined in the 21 children with USCP who did not have borderline (3/50 pronators) or undefined (1/50) pronator muscle activity.

Five of the children had no pronator muscle overactivity during active forearm supination (Fig. 2); 11 of 21 had only one overactive pronator, either the pronator teres ($n = 9$) (Fig. 3) or pronator quadratus ($n = 2$) (Fig. 4), and the remaining 5 children had overactivity of both forearm pronators (Fig. 5).

Four of 7 children with no pronator muscle spasticity according to the MAS had overactivity of 1 or both pronator muscles on EMG during active supination. Two of 11 children with pronator muscle spasticity scores between 1 and 1+ on the MAS had no overactivity on EMG during active supination. Two of 3 children with pronator muscle spasticity scores between 2 and 3 on the MAS had overactivity of only 1 pronator muscle on EMG during active supination.

3.5. Concurrent validity of the surface EMG semi-automatic detection method

To establish the concurrent validity of the surface EMG semi-automatic detection method, it was compared with the visual assessments by the EMG experts, considered the gold standard.

- 23/25 pronator muscles visually determined as overactive by the EMG experts were also found to be excessively active during supination by the surface EMG semi-automatic detection method (3% to 36% of the PS movement cycle), and the remaining 2/25 were not found to be excessively active by the surface EMG semi-automatic detection method.
- 20/21 pronator muscles visually determined as normal by the EMG experts were not found to be excessively active by the surface EMG semi-automatic detection method, and the remaining muscle was found to be excessively active by the surface EMG semi-automatic detection method (1% of the PS movement cycle).
- The 3 pronator muscles visually evaluated as borderline by the EMG experts were found to be excessively active by the surface EMG semi-automatic detection method [1% (1/3), 2% (1/3) and 7% (1/3) of the PS movement cycle].
- The pronator muscle visually evaluated as undefined by the EMG experts was found to be excessively active during 9% of the supination movement by the surface EMG semi-automatic detection method.

From the results of the visual assessment by the EMG experts and the surface EMG semi-automatic detection method, we determined overactivity detection thresholds for the pronator muscles: a muscle was considered:

- overactive when its activation (including variability) during supination was significantly higher ($P < 0.05$) than the mean activation of the TD group (including variability) for at least 3% of the cycle;

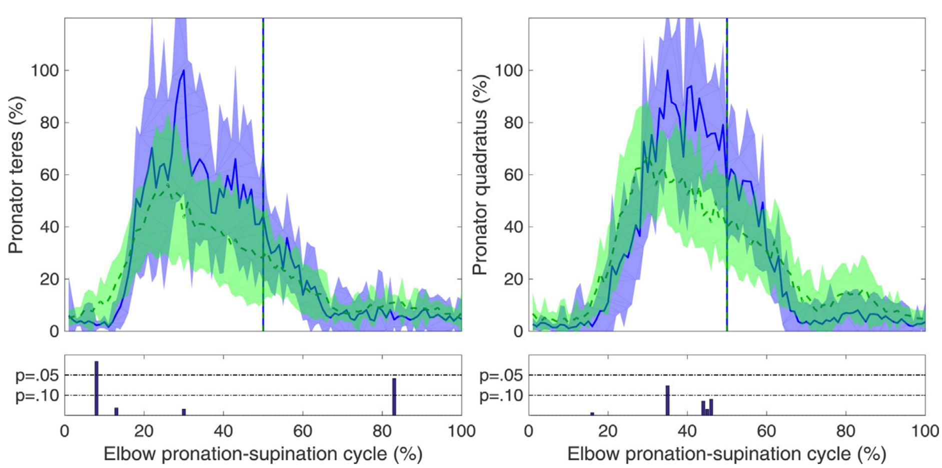

Fig. 2. Mean normalized EMG signal (%) of pronator teres (left) and pronator quadratus (right) muscles of typically developing (TD) children (green dotted line) with its total standard deviation calculated with the inter-trial, inter-session, and inter-subject variability (green shaded area). The EMG signal of child 13 with unilateral spastic cerebral palsy (USCP; blue full line) is shown with its respective total standard deviation calculated with the inter-trial and inter-session variability (blue shaded area) during elbow pronation (0–50%)–supination (50–100%) movements. The statistical significance (p-value) of the deviation is reported for each percentage of the elbow pronation–supination cycle at the bottom of the graph. The vertical line represents the beginning of the supination movement. The EMG experts were unanimous that child 13 did not have overactivity of the pronator teres or pronator quadratus muscle.

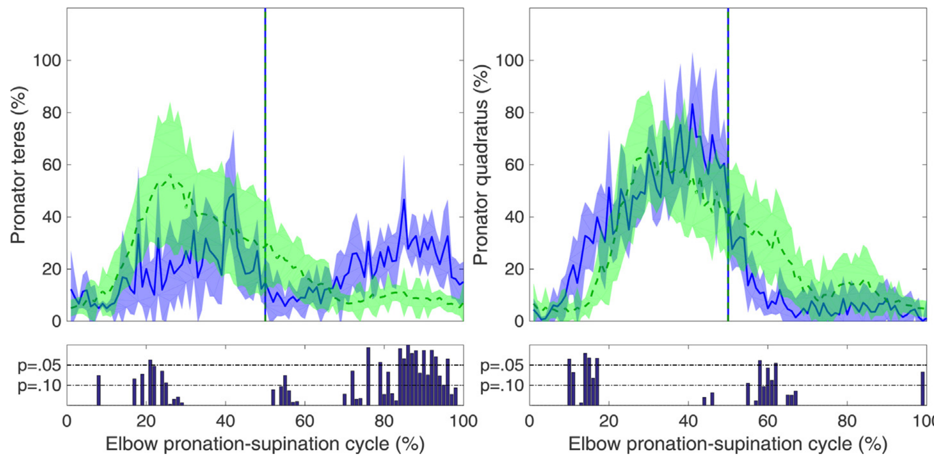

Fig. 3. Mean normalized EMG signal (%) of pronator teres (left) and pronator quadratus (right) muscles of TD children (green dotted line) with its total standard deviation calculated with the inter-trial, inter-session, and inter-subject variability (green shaded area). The EMG signal of child 6 with USCP (blue full line) is shown with its respective total standard deviation calculated with the inter-trial and inter-session variability (blue shaded area) during elbow pronation (0–50%)-supination (50–100%). The statistical significance (*P*-value) of the deviation is reported for each percentage of the elbow pronation–supination cycle at the bottom of the graph. The vertical line represents the beginning of the supination movement. The EMG experts were unanimous that child 6 had overactivity of only the pronator teres muscle.

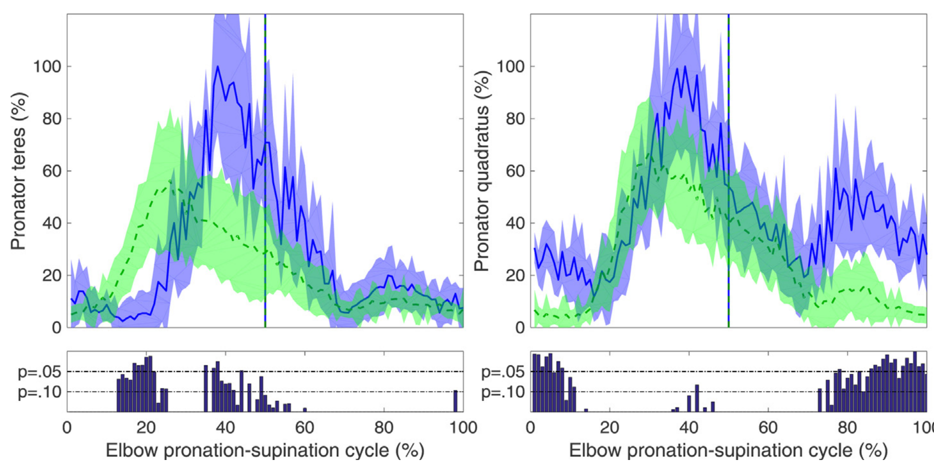

Fig. 4. Mean normalized EMG signal (%) of pronator teres (left) and pronator quadratus (right) muscles of TD children (green dotted line) with its total standard deviation calculated with the inter-trial, inter-session, and inter-subject variability (green shaded area). The EMG signal of child 15 with USCP (blue full line) is shown with its respective total standard deviation calculated with the inter-trial and inter-session variability (blue shaded area) during elbow pronation (0–50%)–supination (50–100%). The statistical significance (p-value) of the deviation is reported for each percentage of the elbow pronation–supination cycle at the bottom of the graph. The vertical line represents the beginning of the supination movement. The EMG experts were unanimous that child 15 had overactivity of only the pronator quadratus muscle.

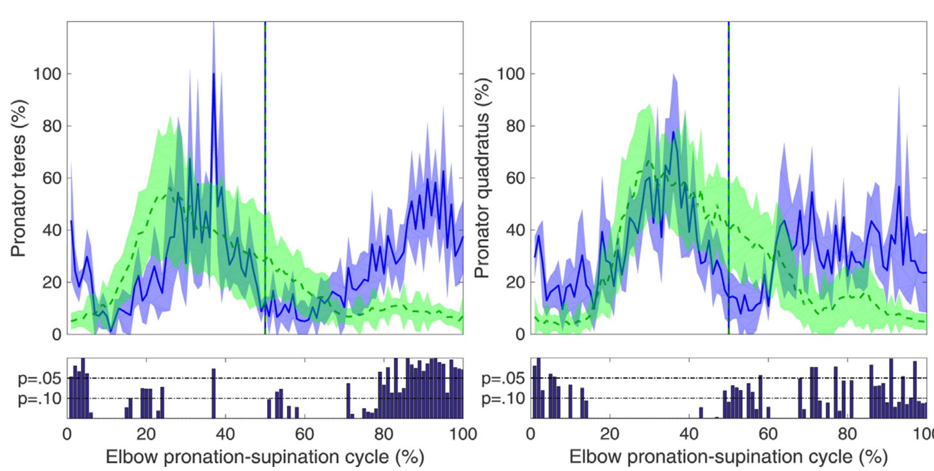

Fig. 5. Mean normalized EMG signal (%) of pronator teres (left) and pronator quadratus (right) muscles of TD children (green dotted line) with its total standard deviation calculated with the inter-trial, inter-session, and inter-subject variability (green shaded area). The EMG signal for child 3 with USCP (blue full line) is shown with its respective total standard deviation calculated with the inter-trial and inter-session variability (blue shaded area) during elbow pronation (0–50%)–supination (50–100%). The statistical significance (*P*-value) of the deviation is reported for each percentage of elbow pronation–supination cycle at the bottom of the graph. The vertical line represents the beginning of the supination movement. EMG experts were unanimous that child 3 had overactivity of both pronator teres and pronator quadratus muscles.

- borderline when its activation was significantly higher for 1% or 2% of the cycle;
- normal when its activation was not significantly higher than that of the reference group.

The use of these thresholds yielded excellent agreement between the surface EMG semi-automatic detection method and the visual assessments by the EMG experts: 45/50 (90%), with Krippendorff's alpha 0.82 (95% CI 0.68–0.97). The positive predictive value was 23/24 (96%) and negative predictive value 20/22 (91%). The borderline pronators were excluded from the calculation of the positive and negative predictive values.

The EMG patterns of both pronator muscles of each child with USCP, their variability and the statistical results are provided in Appendix 2.

4. Discussion

The first objective of this study was to establish whether EMG experts could reliably detect pronator muscle overactivity from post-processed EMG signals. The intra-rater reliability of visual assessment by EMG experts was excellent, but the inter-rater reliability was only moderate. This finding highlights the subjective nature of the clinical interpretation of EMG signals and demonstrates the need for objective methods such as that developed here to assist clinicians in analysing EMG signals.

The second objective was to determine whether EMG experts could discriminate between different pronator-muscle surface EMG activation profiles during active supination movements. Excessive pronator activation was a prevalent feature of the USCP group: 20 of the 25 children evaluated had excessive activation of one or both pronator muscles. All possible profiles of pronator muscle overactivity were found in the sample of 25 children with USCP: no pronator muscle overactivity during active forearm supination; only one overactive pronator (either the pronator teres or pronator quadratus); and overactivity of both forearm pronators. These results provide 2 types of new information (discussed below), which are complementary to the clinical examination.

First, we found no relation between pronator muscle overactivity during active supination and spasticity rated on the MAS, as was previously described [2]. Surface EMG is currently the only non-invasive method of evaluating muscle activation during active movement. This study revealed that some of the children with USCP, despite having severely restricted active supination, had no pronator muscle overactivity, whereas others, who had 1 or even 2 overactive pronator muscles, only had mildly limited active supination. This finding emphasizes that USCP affects each child differently and that the clinical evaluation may be insufficient to understand each child's specific movement impairment. In some children, muscle overactivity is the predominant cause of the movement impairment, whereas in others, paresis is the predominant cause. The information obtained from the dynamic EMG analysis can be used to determine the cause of the movement impairment, and thus an appropriate treatment plan can be established.

Second, the results showed the existence of different pronator-muscle overactivity profiles. This finding is important because it highlights the importance of accurately determining which of the two pronator muscles is overactive before injecting neuromuscular blocking agents to improve supination. From our findings, we suggest that any treatment by a neuromuscular blocking agent should always be preceded by EMG analysis of active supination movements in order to determine which muscles actually require treatment. This would ensure the effectiveness of such treatments, as was suggested previously in adults with hemiparesis [7].

The third objective of this study was to adjust the overactivity detection threshold of the semi-automatic method according to the visual assessment by the EMG experts, to ensure the highest possible concurrent validity of the method. A statistical tool associated with a threshold to distinguish normal activation from excessive antagonist activation is integrated in the surface-EMG semi-automatic detection method. After adjustment of this threshold on the basis of the visual EMG assessments by the experts, the concurrent validity and the positive and negative predictive values of the semi-automatic detection method were excellent (agreement = 90%; Krippendorff's alpha = 0.82 [95% 0.68–0.97]; positive predictive value 96%; negative predictive value 91%). We believe this surface-EMG method of muscle overactivity detection could help clinicians who are inexperienced in the interpretation of surface EMG signals to improve their diagnostic skills.

The surface EMG has some limitations. Surface electrodes may capture signals from more than one muscle, known as crosstalk, especially in the small forearms of children. However, even though the pronator quadratus is a deep muscle, it is isolated in the forearm and its fibres run perpendicular to the forearm, so no other muscle activity can interfere with its signal. The pronator teres muscle is superficial and is surrounded by wrist and finger flexor muscles and thus could be subject to crosstalk. However, of the 25 children with USCP, none had excessive wrist flexion/extension movements during supination, and 3 had slightly excessive wrist radial deviation during supination. Those children had different pronator-muscle overactivity profiles; in particular, the child with the largest radial deviation had no pronator overactivity. This finding suggests that little crosstalk occurred between the pronator teres signal and surrounding muscles during the PS movements. Intramuscular EMG could eliminate any possibility of crosstalk but is invasive and is associated with other issues such as pain caused by the fine wires that can limit active movement and the possible need for drugs or gas to relax the child during insertion of the wires.

Another limitation of this study is the EMG amplitude normalization method. EMG amplitude was normalized by the maximal amplitude recorded during elbow movements. If the force generated by the muscle was relatively low during these movements (due to the paresis) then the degree of muscle activation may have been overestimated (relative to the activation recorded in the reference group), thereby leading to an overestimation of overactivity. However, this limitation is unavoidable because of currently no method of EMG amplitude normalization that can take paresis into account [18,32]. Furthermore, overestimation can be amplified when using normalisation methods that require high levels of muscle force such as Maximal Voluntary Contractions.

Despite these limitations, the surface-EMG semi-automatic detection method proposed here provides new and complementary information for the assessment of motor impairment in children with USCP. Although this study was conducted in a motion analysis laboratory, a complex structure rarely found in rehabilitation centres outside university hospitals, the method can also be applied to EMG signals recorded using simple, portable surface EMG/video systems available in most rehabilitation centres.

5. Conclusions

This study aimed to develop a method to detect pronator muscle overactivity during active supination in children with USCP by using surface EMG. The method had a positive predictive value of 96% and negative predictive value of 91%, based on visual assessment by EMG experts. This method could help inexperienced clinicians interpret EMG signals from pronator muscles. However, users must be aware of the limitations of surface EMG, in particular the risk of crosstalk and the fact that paresis cannot be detected by the EMG signals.

The results revealed different profiles of pronator muscle overactivity present in a sample of 25 children with USCP. Surface-EMG analysis provides a complement to the typical clinical examination of children with CP. In light of our findings, we recommend that the decision to inject a neuromuscular blocking agent be preceded by a simple EMG analysis of active supination movements to precisely determine which muscles are overactive and which are not.

Future studies should evaluate the use of this method for other movements and agonist muscle groups.

Disclosure of interest

The authors declare that they have no competing interest.

Acknowledgements

We are grateful to the children and their parents for their collaboration. We thank Johanna Robertson and Jennifer Dandrea-Palethorpe for their thorough and precise English proofreading. We thank external EMG expert François Hug for his time spent analysing the EMG signals and his precious advice. This work was supported by grants from the Fondation Paralysie Cérébrale and Envoludia.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at https://doi.org/10.1016/j.rehab.2019.08.001.

References

[1] Levitt S. Treatment of cerebral palsy and motor delay, 5th, Chichester, UK: Wiley-Blackwell; 2010.
[2] Gracies J-M. Pathophysiology of spastic paresis. II: emergence of muscle overactivity. Muscle Nerve 2005;31:552–71.
[3] Unnithan VB, Dowling JJ, Frost G, Volpe Ayub B, Bar-Or O. Cocontraction and phasic activity during GAIT in children with cerebral palsy. Electromyogr Clin Neurophysiol 1996;36:487–94.
[4] Ikeda AJ, Abel MF, Granata KP, Damiano DL. Quantification of cocontraction in spastic cerebral palsy. Electromyogr Clin Neurophysiol 1998;38:497–504.
[5] Bohannon RW, Smith MB. Interrater reliability of a modified Ashworth scale of muscle spasticity. Phys Ther 1987;67:206–7.
[6] Boyd RN, Graham HK. Objective measurement of clinical findings in the use of botulinum toxin type A for the management of children with cerebral palsy. Eur J Neurol 1999;6:s23–35.
[7] Gracies J-M, et al. Safety and efficacy of abobotulinumtoxin A for hemiparesis in adults with upper limb spasticity after stroke or traumatic brain injury: a double-blind randomised controlled trial. Lancet Neurol 2015;14:992–1001.
[8] Rameckers EAA, Speth LAWM, Duysens J, Vles JSH, Smits-Engelsman BCM. Botulinum toxin-a in children with congenital spastic hemiplegia does not improve upper extremity motor-related function over rehabilitation alone: a randomized controlled trial. Neurorehabil Neural Repair 2009;23:218–25.
[9] Gross R, Leboeuf F, Hardouin JB, Perrouin-Verbe B, Brochard S, Rémy-Néris O. Does muscle coactivation influence joint excursions during gait in children with and without hemiplegic cerebral palsy? Relationship between muscle coactivation and joint kinematics. Clin Biomech 2015;30:1088–93.
[10] Gross R, et al. The influence of gait speed on co-activation in unilateral spastic cerebral palsy children. Clin Biomech 2013;28:312–7.
[11] Damiano DL, Martellotta TL, Sullivan DJ, Granata KP, Abel MF. Muscle force production and functional performance in spastic cerebral palsy: relationship of cocontraction. Arch Phys Med Rehabil 2000;81:895–900.
[12] Sarcher A, Raison M, Leboeuf F, Perrouin-Verbe B, Brochard S, Gross R. Pathological and physiological muscle co-activation during active elbow extension in children with unilateral cerebral palsy. Clin Neurophysiol 2017;128:4–13.
[13] Sarcher A, et al. Impact of muscle activation on ranges of motion during active elbow movement in children with spastic hemiplegic cerebral palsy. Clin Biomech 2015;30:86–94.
[14] Malfait N, Sanger TD. Does dystonia always include co-contraction? A study of unconstrained reaching in children with primary and secondary dystonia. Exp Brain Res 2007;176:206–16.
[15] de Bruin M, Veeger HEJ, Kreulen M, Smeulders MJC, Bus SA. Biceps brachii can add to performance of tasks requiring supination in cerebral palsy patients. J Electromyogr Kinesiol 2013;23:516–22.
[16] Steenbergen B, Gordon AM. Activity limitation in hemiplegic cerebral palsy: evidence for disorders in motor planning. Dev Med Child Neurol 2006;48:780.
[17] Kreulen M, Smeulders MJC, Veeger HEJ, Hage JJ. Movement patterns of the upper extremity and trunk associated with impaired forearm rotation in patients with hemiplegic cerebral palsy compared to healthy controls. Gait Posture 2007;25:485–92.
[18] Sarcher A, et al. Patterns of upper limb muscle activation in children with unilateral spastic cerebral palsy: variability and detection of deviations. Clin Biomech 2018;59:85–93.
[19] Schwartz MH, Trost JP, Wervey RA. Measurement and management of errors in quantitative gait data. Gait Posture 2004;20:196–203.
[20] Chia K, Sangeux M. Quantifying sources of variability in gait analysis. Gait Posture 2017;56:68–75.
[21] Eliasson A-C, et al. The Manual Ability Classification System (MACS) for children with cerebral palsy: scale development and evidence of validity and reliability. Dev Med Child Neurol 2006;48:549–54.
[22] Laitenberger M, Raison M, Périé D, Begon M. Refinement of the upper limb joint kinematics and dynamics using a subject-specific closed-loop forearm model. Multibody Syst Dyn 2015;33:413–38.
[23] Malanga GA, Campagnolo DI. Clarification of the pronator reflex. Am J Phys Med Rehabil 1994;73:338–40.
[24] Remaley D, et al. Surface electromyography of the forearm musculature during the windmill softball pitch. Orthop J Sport Med 2015;3:1–8.
[25] Basmajian JV. Primary Anatomy, 8th, Baltimore: Williams and Wilkins; 1982.
[26] Barre A, Armand S. Biomechanical ToolKit: open-source framework to visualize and process biomechanical data. Comput Methods Programs Biomed 2014;114:80–7.
[27] Lu T-W, O'Connor JJ. Bone position estimation from skin marker co-ordinates using global optimisation with joint constraints. J Biomech 1999;32:129–34.
[28] Hodges PW, Bui BH. A comparison of computer-based methods for the determination of onset of muscle contraction using electromyography. Electroencephalogr Clin Neurophysiol 1996;101:511–9.
[29] Krippendorff K. Computing Krippendorff's alpha-reliability. Dep Pap 2011 [Retrieved from http://repository.upenn.edu/asc_papers/43].
[30] Krippendorff K. Reliability in content analysis. Hum Commun Res 2004;30:411–33.
[31] Lydersen S, Pradhan V, Senchaudhuri P, Laake P. Choice of test for association in small sample unordered $r \times c$ tables. Stat Med 2007;26:4328–43.
[32] Hug F. Can muscle coordination be precisely studied by surface electromyography? J Electromyogr Kinesiol 2011;21:1–12.