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# Cervical dystonia: Improved treatment response to botulinum toxin after referral to a tertiary centre and the use of polymyography

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ABSTRACT

*Rationale:* Cervical dystonia is the most common form of (primary) dystonia. The first line of treatment for cervical dystonia is intramuscular injections with botulinum toxin. To optimise the response to botulinum toxin proper muscles selection is required. Pre-treatment polymyographic EMG in addition to clinical evaluation is hypothesised to be a good tool to improve muscle selection and treatment outcome.
*Objective:* To determine the efficacy of botulinum toxin treatment after adjacent polymyographic EMG in cervical dystonia patients referred to our tertiary referral centre with an unsatisfactory response to botulinum toxin treatment elsewhere.
*Methods:* We performed a retrospective analysis of 40 consecutive second opinion cervical dystonia patients. Standard polymyographic EMG was performed before treatment. We retrieved the Tsui scores and subjective evaluations from the first visit, after 12 weeks and after one year of treatment. In addition, we assessed the final outcome of treatment in our centre based on the records and asked the patients for their personal opinion about the effect of referral to our centre on their treatment response.
*Results:* After one year of treatment there was a significant improvement on both the Tsui scores ($p < 0.01$) and the subjective treatment evaluation ($p < 0.001$.) On their last visit 60% of the patients still continued treatment with a reasonable to good response.
*Conclusion:* A substantial amount of CD patients with an unsatisfactory response to botulinum toxin improved after polymyography and subsequent treatment with botulinum toxin in a tertiary referral centre.



## 1. Introduction

Dystonia is a syndrome characterized by sustained involuntary muscle contractions leading to debilitating abnormal postures, pain and twisting movements. Cervical dystonia (CD), also called spasmodic torticollis, is the most common form of (primary) dystonia [1–3]. In simple rotational torticollis, the most frequent dystonic posture in CD, the sternocleidomastoid (SCM), the splenius capitis (SPL) and the obliquus capitis (OC) muscles are typically involved. In more complex movements, such as laterocollis, antecollis, retrocollis, and tremulous torticollis, other combinations of muscles are active [4–6].

The first line of treatment for CD is intramuscular injections with botulinum toxin (BoNT) in the dystonic muscles. BoNT treatment results in significant improvement of resting posture and reduction of pain in about 85% of the patients [7,8].

The supposedly most important determinants of a favourable response to BoNT treatment include proper selection of dystonic muscles and an appropriate dosage of BoNT [9]. The selection of muscles to be injected with BoNT is usually based on clinical features, such as abnormal posture, muscle palpation, muscle hypertrophy, and pain. In addition to clinical examination, polymyographic electromyography (*p*EMG) can be used to determine the pattern of muscle involvement. Some studies indicate that *p*EMG for muscle selection may help to improve the response to botulinum toxin treatment, but evidence is scarce [10]. The Academic Medical Center (AMC) is a national reference centre for patients with dystonia in the Netherlands. Many CD patients with an unsatisfactory response to botulinum toxin treatment are referred to our outpatient clinic. Before treatment, all patients undergo *p*EMG. Based on the clinical evaluation combined with *p*EMG results patients are treated with botulinum toxin injections. To determine the efficacy of referral to our tertiary referral centre

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and investigate the additional value of *p*EMG, a retrospective analysis was performed of CD patients referred with an unsatisfactory response to botulinum toxin treatment.

## 2. Methods

Forty consecutive patients with idiopathic CD referred to our specialized centre between 2003 and 2009 were studied. The following inclusion criteria were chosen: 1) diagnosis of CD, 2) referred because of unsatisfactory response to botulinum toxin treatment and 3) *p*EMG performed in our centre before treatment. Exclusion criteria were: 1) an expressed suspicion in our centre of a psychogenic cause for the torticollis and 2) insensitivity to botulinum toxin confirmed by a negative extensor digitorum brevis (EDB) test [11].

To obtain the relevant information, patient charts and all the available correspondence were studied. Information was obtained about disease duration, number of previous treatments, muscles selected for treatment, dosages in the referring hospital and in our centre, performance of *p*EMG in the referring hospital and side effects. If patients received Botox mouse-units (MU), the equivalent in Dysport-units was calculated using a conversion ration of 1:3. The number of treatments in the referring hospital was estimated assuming an average of 4 treatments per year if the exact number was unknown.

As outcome measures we retrieved Tsui scores (an objective severity scale ranging from 0 to 25 points [12]), VAS scores (a subjective ten point severity scale) and patients subjective evaluations assessed at the first visit in our centre, after 12 weeks and after one year of treatment. Patient's subjective evaluations reflected the perceived effect of the previously received treatment and were scored by one of the investigators (S.N.) as good effect (1), moderate effect (2) or no effect (3). In addition, we assessed the final treatment outcome in our centre based on the last entries in the patient charts. The final outcome was scored as follows: continuing of botulinum toxin treatment in our centre or with their referring neurologist with a good (1), reasonable (2) or poor (3) response, referral to a neurosurgeon for Deep Brain Stimulation (DBS) (4), withdrawal from treatment (5) or unknown (6). In case of ongoing treatment (1–3) the general treatment response was based on the average subjective evaluations from the last treatments. A treatment response was qualified as good (1) if there was a good treatment response in the vast majority of the treatments. A treatment response was qualified as reasonable (2) when response was good in at least half of the treatments and treatment response was qualified as poor (3) when the response was moderate or poor in the majority of the treatments. Finally, we attempted to reach all patients to ask their final personal opinion about the overall effect of referral to our centre on their treatment response. Their response was scored as clear improvement (1), some improvement (2), no improvement (3) or worsening (4) of treatment response.

### 2.1. Treatment protocol

Before the first botulinum toxin treatment in our centre *p*EMG was performed, using needle electrode recordings with auditory and visual feedback. Based on clinical evaluation combined with *p*EMG results, dystonic muscles were selected for BoNT treatment. All patients received BoNT every three months. A dilution of 200 MU dysport per millilitre was used and injections were divided over 1–3 injection sites per muscle. During all treatment sessions EMG guidance with auditory/visual feedback was used for accurate needle placement in target muscles. All patients were treated by an experienced team consisting of 3 movement disorder specialists and 2 specialized dystonia nurse practitioners. Treatment protocols and injections schedules were always determined by a movement disorder specialist. Before each subsequent treatment patients were asked for side effects experienced.

## 3. Statistical analysis

All statistics were performed using SPSS 16.0. Baseline characteristics are presented using mean and SD for normally distributed data and median and inter quartile range (IQR) for not normally distributed data. The Wilcoxon Signed Ranks Test was used to compare the mean Botulinum toxin dose patients received before referral with the dose given at the first and at the last treatment session in our centre. Longitudinal analyzes were used to investigate the improvement of the Tsui and subjective scores. For the longitudinal analyzes only patients were included with data on all the time points. The Wilcoxon Signed Ranks Test was used to compare the subjective scores before treatment to the scores after one year of treatment and the paired sample *t* test was used to compare the Tsui scores. To investigate the association between the use of *p*EMG in the referring hospital and a favourable outcome in our centre the Pearson's chi squared test and the Fisher exact test were used. A favourable outcome was defined as a reasonable or good final treatment outcome or a clear subjective overall improvement. Finally, to compare the proportions of primary non-responders and secondary non-responders with a favourable outcome, the Pearson's chi squared test was used.

## 4. Results

Forty three consecutively referred patients matching all the inclusion criteria were identified. Two patients had a negative EDB test and were excluded. One patient died and his patient chart could not be retrieved, so he was also excluded. The remaining 40 patients were included for analysis. Baseline characteristics are presented in Table 1.

## 5. Dosage of botulinum toxin

The dosage of botulinum toxin received during the last treatment in the referring centre could be found for only 21 cases. Ten patients received injections with Botox and 11 with Dysport with a median (converted) dose of 375 MU(IQR 285–500). The difference between the last dosage at the referring centre (375 units) and the first dosage in our centre (median 355 units) was not statistically significant ($p > 0.05$). The dosage significantly increased gradually from 355 units (IQR 282–450) at the first treatment in our centre to 400 units (IQR 350–470) after one year ($p < 0.05$). However, the last dosage received at the referring centre and the dosage after one year of treatment in our centre did not differ significantly ($p > 0.05$) (Fig. 1).

## 6. Muscle selection

In 25 of the 40 (63%) patients the selection of muscles for treatment in the referring centre could be retrieved. In 24 of these 25 patients (96%) the selection of muscles changed after *p*EMG evaluation and clinical evaluation in our centre. The median number of mutations in muscle selection was 4 (IQR 3–5). The median number of additional muscles that were injected was 2 (IQR 1.5–3.5) and the median number of muscles that were no longer treated after *p*EMG evaluation was 1 (IQR 1–2). The muscles that were most frequently added to the muscles selected for treatment were the splenius (SPL) and the semispinalis (SESP) muscles. The levator scapulae (LS) and the trapezius (TPZ) muscles were most frequently removed from the selection.

## 7. Tsui, VAS and subjective scores

The mean Tsui score at time of referral was 11.2 ($N = 36$). After one year 29 patients still continued BoNT treatment in our centre. After excluding cases with missing values, longitudinal analysis was performed on the Tsui scores ($N = 24$) and on the subjective scores ($N = 23$). The mean Tsui score in these patients improved from a baseline score of 11.2 at time of referral to 10.3 after 12 weeks (8% improvement). After one year of treatment in our centre the Tsui

**Table 1**
Baseline characteristics.

| | |
|---|---|
| Male | 14/40 (35%) |
| Age | 56 years (mean, SD: 11.9) |
| Disease duration | 7.5 years (median, IQR: 2.25–13.75) |
| Number of previous treatments | 5 (median, IQR: 2–14) |
| Baseline Tsui (0–25 points) | 11.2 (mean, SD: 3.1) |
| Baseline VAS (0–10 points) | 8.2 (mean, SD: 1.15) |
| Dosage (last in referring centre) | 375 MU (median, IQR 285–500) |
| Primary non-responders | 69% |

SD: standard deviation. IQR: inter quartile range.

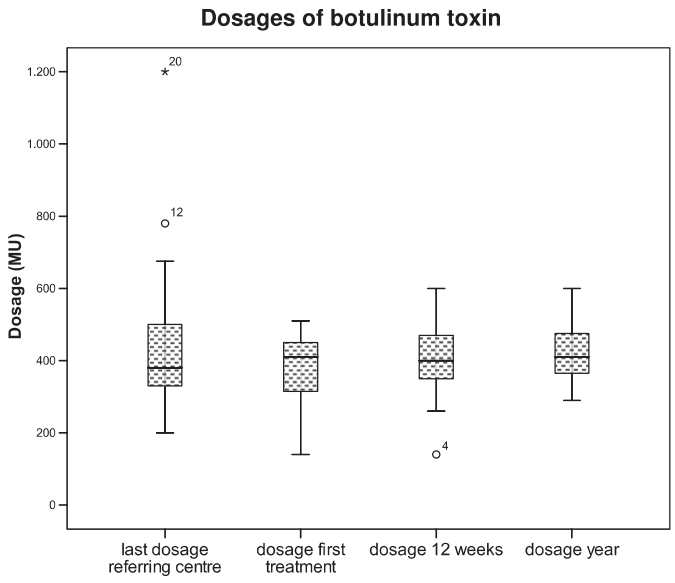


**Fig. 1.** Dosage. Ns: not significant ($p > 0.05$) *: statistically significant ($p < 0.05$) O: outlier * extreme outlier.

scores had significantly improved to 9.1 (18.8% improvement, $p < 0.01$). The subjective scores significantly improved after 12 weeks ($p < 0.01$) and even further after one year ($p < 0.001$) of treatment (Fig. 2). Longitudinal analysis could not be performed on the VAS scores as for only a small number of patients data was available on all time points (Table 2).

## 8. Final treatment outcome

On their last visit, on average 2.4 year (range 0.5–6) after the first treatment in our centre, 32.5% of the patients still continued botulinum toxin treatment with a good response, 27.5% continued with a reasonable response and 2.5% continued with a poor response. Twenty percent of the patients withdrew from treatment, 15% were referred for Deep Brain Stimulation (DBS) and for one patient the treatment outcome was unknown.

Thirty-four patients (85%) could be reached to ask their personal final opinion about the effect of referral on their treatment response. Seventeen patients (50%) thought that their response to botulinum toxin treatment improved and 11 of these 17 patients reported a clear improvement. Sixteen patients (47%) thought there was no difference in treatment response after continuing treatment in our centre and one patient thought that his response had decreased.

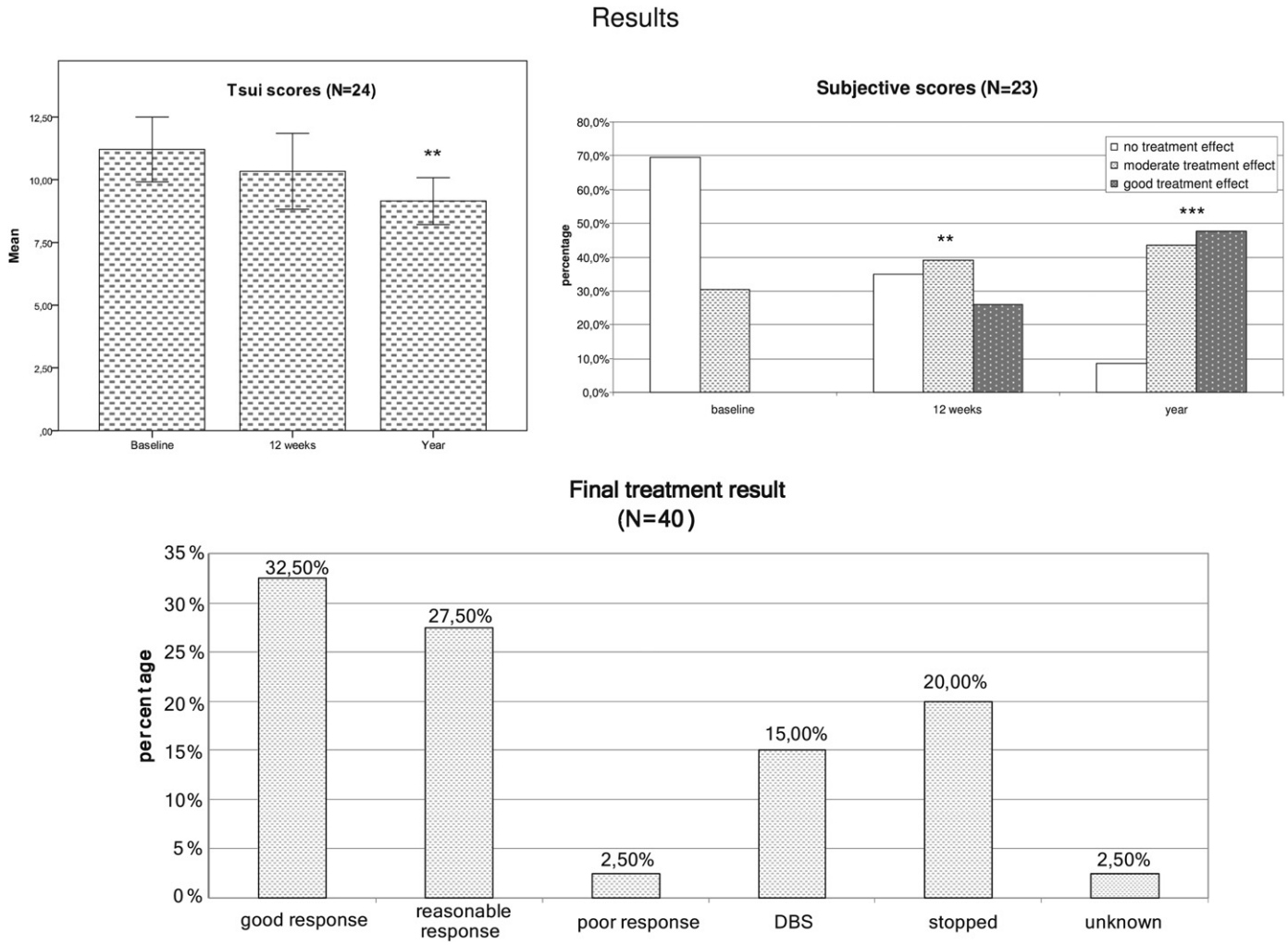


**Fig. 2.** Results. **: Significantly different from baseline score ($p$-value $< 0.01$), ***: significantly different from baseline score ($p$-value $< 0.001$) error bars: 95% confidence interval. DBS: Deep brain stimulation.

**Table 2**
Results.

| Pat nr | Predominant movement | Primary/ secondary non-responders | Age | Duration disease (years) | Number of previous treatments | Last dosage referring centre (MU) | Previous polymyography | Tsui | | | Visual analogue scale | | | Subjective scores[a] | | | Final treatment outcome[b] | Overall subjective evaluation[c] | First dose (MU) | Second 12 wks (MU) | Dose year (MU) |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | | | | Baseline | 12 wks | Year | Baseline | 12 wks | Year | Baseline | 12 wks | Year | | | | | |
| 1 | Antecollis | P | 65 | 10 | 20 | | | | 8 | | | | | 3 | 3 | | IV | 3 | 140 | 260 | |
| 2 | Rotation (r) | | 67 | 3 | 16 | | Y | 10 | 12 | 9 | | 4 | 6 | | 2 | 1 | I | 3 | 400 | 360 | 360 |
| 3 | Rotation (l)/ lateroflexion (r) | P | 60 | 15 | 2 | | | 8 | 12 | 8 | | 8 | | 2 | 3 | 3 | V | | 490 | 710 | 700 |
| 4 | Rotation (r) | P | 66 | 11 | 20 | 270 | N | | 12 | 9 | | | | 3 | 2 | 1 | I | 1 | 140 | 140 | 340 |
| 5 | Rotation (r) | S | 50 | 10 | 2 | 500 | | 12 | 12 | 12 | 9 | | | 3 | 1 | 1 | I | | 480 | 520 | 560 |
| 6 | Lateroflexion (L) | P | 47 | 2 | 2 | 200 | N | 14 | 10 | 14 | | | | 3 | 3 | 2 | I | 1 | 260 | 370 | 380 |
| 7 | Rotation (l) | P | 59 | 5 | 5 | 380 | N | | 13 | | | 9 | 8 | 3 | 3 | 2 | II | 1 | 400 | 480 | 480 |
| 8 | Retrocollis | S | 48 | 14 | 36 | | N | 16 | 16 | 7 | 8 | 8 | | | 2 | 1 | II | 1 | 330 | 350 | 400 |
| 9 | Rotation (l) | S | 44 | 13 | 20 | 675 | Y | 15 | 12 | 12 | 9 | 10 | 10 | 3 | 2 | 1 | II | 2 | 450 | 280 | 400 |
| 10 | Rotation (r)/ lateroflexion (r) | S | 60 | 28 | 12 | | | 11 | 8 | 8 | | | 8 | 2 | 3 | 3 | I | | 270 | 320 | 400 |
| 11 | Rotation (r) | P | 42 | 1.5 | 2 | 500 | | 10 | 6 | 8 | 8 | 8 | 6 | 3 | 1 | 1 | II | | 450 | 390 | 490 |
| 12 | Lateroflexion (r) | P | 50 | 7 | 2 | 780 | N | 18 | 20 | 10 | | | 9 | 3 | 3 | 2 | I | 1 | 300 | 600 | 410 |
| 13 | Rotation (r)/ lateroflexion (l) | P | 65 | 2 | 5 | 140 | Y | 8 | 8 | | | 10 | | 3 | 3 | | V | 3 | 260 | 200 | |
| 14 | Rotation(r) | P | 28 | 2 | 2 | | N | 14 | 12 | 7 | 9 | | | 3 | 2 | 1 | I | 1 | 370 | 490 | 460 |
| 15 | Rotation (l) | S | 48 | 0.75 | 2 | 450 | N | 10 | 10 | 12 | 7 | | | 2 | 3 | | I | 1 | 390 | 370 | 460 |
| 16 | Rotation(r)/ retroflexion | P | 67 | 3 | 3 | 360 | N | 6 | 10 | | 7 | 8 | | 3 | 2 | 2 | V | 3 | 410 | 430 | 430 |
| 17 | Rotation (r) | P | 48 | 18 | | | N | 5 | 6 | 6 | 7 | | | 3 | 2 | | II | 2 | 200 | 280 | 350 |
| 18 | Rotation (l)/ lateroflexion(r) | P | 39 | 1 | 2 | 400 | Y | 14 | | | | | | 3 | | | V | 3 | 350 | | |
| 19 | Rotation (l) | S | 54 | 2 | 8 | 375 | N | 8 | 4 | 4 | | | 5 | 2 | 1 | 1 | II | 1 | 420 | 460 | 370 |
| 20 | Rot (l) lat (r) | P | 52 | 15 | | 1200 | N | 8 | 6 | 8 | | | | 3 | 3 | 1 | I | 1 | 450 | 450 | 470 |
| 21 | Rotation (l) | P | 75 | 30 | 3 | 300 | | 12 | 8 | 9 | | 7 | 6 | 3 | 1 | 2 | III | 3 | 250 | 260 | 290 |
| 22 | Rotation (l) | P | 75 | 8 | 24 | | | 16 | 18 | | 9.5 | 9 | | 2 | 3 | | IV | 3 | 490 | 680 | |
| 23 | Rotation (l) | P | 74 | 2 | 2 | 500 | N | 10 | 8 | 10 | 7.5 | | 8 | 3 | 3 | 1 | I | 1 | 510 | 570 | 600 |
| 24 | Rotation (l) | S | 65 | 12 | 48 | 450 | | 11 | 7 | | 6.5 | | | 2 | 2 | | II | 3 | 350 | 350 | |
| 25 | Rotation (r)/ anteroflexion | P | 63 | 3 | 6 | | N | 12 | 7 | 8.5 | 7.5 | | | 2 | 2 | 2 | IV | 3 | 470 | 370 | 240 |
| 26 | Rotation (r) | S | 61 | 4 | 9 | 250 | N | 12 | 9 | 12 | | | | 2 | 1 | 2 | II | 2 | 330 | 330 | 340 |
| 27 | Rotation (r) | P | 33 | 10 | 3 | | | 11 | | | | | | 2 | | | VI | | 400 | | |
| 28 | Anteroflexion | S | 55 | 15 | 20 | 165 | | 12 | 12 | | | | | 2 | 2 | | V | 3 | 300 | 260 | |
| 29 | Rotation (l) | | 42 | 15 | 12 | 330 | Y | | | | | | | 2 | | | IV | 4 | 360 | | |
| 30 | Rotation (l) | S | 59 | 1.5 | 6 | 320 | N | 8 | 8 | | | | | 2 | 1 | | II | 2 | 320 | 440 | |
| 31 | Retroflexion | P | 64 | 4 | 2 | | Y | 12 | 12 | 10 | | | | 3 | 3 | 1 | II | 2 | 200 | 280 | 400 |
| 32 | Rotation (l)/ lateroflexion (r) | P | 74 | 5 | 2 | | N | 14 | 14 | 9 | 8 | 9 | | 3 | 2 | 2 | I | | 490 | 600 | 580 |
| 33 | Rotation (l)/ lateflexion (l) | S | 50 | 10 | 36 | | N | 10 | 12 | 10 | 10 | 8 | 10 | 3 | 2 | 2 | IV | 3 | 450 | 420 | 360 |
| 34 | Rotation (l) | S | 51 | 2 | 2 | | N | 16 | 12 | | 10 | | | 3 | 3 | | IV | 3 | 380 | 410 | |
| 35 | Rotation (l)/ lateroflexion (r) | P | 68 | 10 | 7 | | | 12 | | | | | | 2 | 2 | | V | 3 | 340 | | |
| 36 | Rotation (r)/ lateroflexion (l) | | 42 | 9 | 2 | 360 | N | 10 | 9 | 8 | | 4 | 8 | 2 | 2 | 1 | II | 1 | 500 | 400 | 360 |
| 37 | Anteroflexion | P | 72 | 4 | 4 | | N | 12 | 10 | 10 | | 7 | 3 | 3 | 2 | 2 | I | 3 | 240 | 300 | 300 |
| 38 | Rotation (l) | P | 48 | 3 | 4 | | Y | 6 | 13 | 8 | | | 8 | 2 | 1 | 1 | I | 2 | 280 | 310 | 350 |
| 39 | Lateroflexion (L) | P | 56 | 19 | | | Y | 13 | 7 | | | | | 3 | 2 | | V | 3 | 310 | 380 | |
| 40 | Retroflexion | S | 68 | 30 | 8 | | | 7 | 9 | | | | | 3 | 2 | | V | 3 | 290 | 320 | |
| | Average | | 56 | 9.0 | 9.7 | 424 | | 11.2 | 10.3 | 9.1 | 8.2 | 7.8 | 7.3 | 2.6 | 2.2 | 1.6 | | | 355.5 | 392.8 | 417.8 |

MU: Mouse-units, wks: weeks, (r): right, (l): left.

[a] Subjective scores: 1: good effect, 2: moderate effect, 3: no effect.

[b] Roman numerals: I Good response, II reasonable response, III poor response, IV DBS Deep Brain Stimulation, V quit treatment, VI unknown.

[c] Numerals overall subjective evaluation:1 clear improvement, 2 some improvement, 3 no improvement, 4 deterioration.

## 9. Previous use of *p*EMG

Information about the use of *p*EMG in the referring centre was available for 27 patients. Eight of these 27 underwent *p*EMG before treatment in the referring centre whereas 19 patients did not undergo *p*EMG. The eight patients that underwent *p*EMG in the referring centre had relatively little benefit from referral to our centre. The proportion of patients with a 'good' to 'reasonable' final treatment result was smaller in this group (Odds Ratio: 0.267, $p > 0.05$). Similarly, a significantly smaller proportion of this group subjectively reported a clear improvement after referral to our centre ($p < 0.01$) compared to the patients that did not undergo *p*EMG in their referring centre.

## 10. Primary vs secondary non-responders

The proportion of primary non-responders with a good or reasonable final treatment result was slightly smaller compared to secondary non-responders (OR: 0.44 ($p > 0.05$)). However, a slightly bigger proportion of the primary non-responders subjectively reported a clear improvement after referral to our centre (OR: 1.88 ($p > 0.05$)). These differences were not statistically significant.

## 11. Side effects

After the first treatment in our centre 17 patients (42.5%) experienced side effects. Eight patients (20%) complained of pain, 5 patients experienced symptoms of mild dysphagia (12.5%), 5 patients complained of muscle weakness (12.5%) and 2 patients (5%) experienced another side effect (flu-like symptoms in one patient and difficulty to open his mouth in another patient). All side effects were mild, resolved spontaneously and additional interventions were never required. No data were available for side effects from the referring hospitals.

During the following year of treatment in our centre one patient had an exacerbation of pre-existing swallowing difficulties after botulinum toxin treatment and was admitted for 2 days at our hospital for evaluation. No additional interventions, like enteral nutrition, were needed and he left the hospital in a good condition.

## 12. Discussion

A substantial amount of CD dystonia patients with an unsatisfactory response to botulinum toxin treatment improved after polymyography and subsequent botulinum toxin treatment in our centre. A recent review has shown that polymyographic analysis of muscle involvement in CD might improve the response to botulinum toxin treatment in CD, especially in more complex cases, but evidence is scarce [10]. Cordivari et al. [13] have shown that *p*EMG might restore responsiveness in secondary non-responders without antibodies against BoNT.

In our study we were able to obtain a reasonable to good final treatment result in 60% of the patients with a previous unsatisfactory response. In line with this, 50% of the patients reported subjectively, that referral to our centre improved their response to botulinum toxin treatment. The Tsui and subjective scores both improved after a single treatment session, and even further after one year of treatment. Interestingly, the small proportion of patients that underwent *p*EMG already before coming to our centre had relatively little benefit from referral to our centre, supporting the hypothesis of a positive value of additional *p*EMG. The influence of the expertise of our tertiary referral centre on the improved treatment response cannot be ruled out. Also, the influence of EMG guidance for accurate needle placement during BoNT injections can partly explain the positive effect. Finally, the increasing dosage might be explanatory for some of the observed improvement response after one year. However, the increasing treatment response started already after the first treatment in our centre when a lower dose was given compared to the referring centre. There are some limitations to our retrospective study. Eleven of the forty patients were treated for less then one year in our centre and therefore no data were available for all the time points. A substantial part of these eleven patients stopped treatment or were referred for DBS. This is a source of bias towards a favourable outcome of the per protocol analysis that was performed to investigate the improvement on the Tsui and subjective scores after one year of treatment. However, the observed improvement in the remaining patients signifies that a substantial part of the initial non-responders had clearly improved.

In addition there were some missing data, especially in the VAS scores, that could not be retrieved because they were not recorded in the patient charts. We expect these missing data to be rather random and not a substantial source of bias.

Another possible confounding factor might be the time interval between the assessment of the baseline scores and the last treatment patients had received at the referring centre which was often longer than the standard 12 weeks interval between treatment and evaluation of the treatment in our centre. This may have influenced the baseline Tsui scores since they evaluate the severity of the complaints at the time of assessment and this could have biased the observed improvement on these scores. However, subjective scores also indicated a more favourable response in our centre compared to earlier treatment. Finally, some patients were previously treated with Botox and received Dysport in our centre. Although unlikely, as they are both type A forms of botulinum toxin, it cannot be excluded that this switch accounts for part of the favourable response in some patients. This has previously been suggested for patients suffering from blepharospasm and hemificial spasm [14].

Despite the above mentioned limitations, the results are in line and indicate an improved treatment response in a tertiary centre in a substantial part of the patients. We speculate that *p*EMG is helpful in muscle selection and improves the response to botulinum toxin treatment in CD patients. It has been suggested that in the majority of CD patients a good clinical response can be obtained with clinical evaluation alone and in these patients the use of *p*EMG cannot be justified [15]. We hypothesise that even in patients with a good response, treatment results can be further improved with *p*EMG. However, there is insufficient evidence to recommend the standard use of pEMG in all CD patients. Nevertheless, in patients with an unsatisfactory response, inadequate muscle selection might be an important cause for treatment failure and for these patients we recommend the use of *p*EMG to improve treatment response. More prospective randomized trials are required to confirm the value of *p*EMG.

## Conflicts of interest and financial disclosure

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S.W.R. Nijmeijer, T.S.M. Standaar and M. Postma: none.

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