Parkinsonism and Related Disorders 23 (2016) 72–79

Contents lists available at ScienceDirect

Parkinsonism and Related Disorders

journal homepage: www.elsevier.com/locate/parkreldis

A randomized trial of specialized versus standard neck physiotherapy in cervical dystonia

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ARTICLE INFO

Article history:
Received 3 August 2015
Received in revised form
24 November 2015
Accepted 15 December 2015

Keywords:
Torticollis
Physical therapy modalities
Single-blind method
Disability
Evaluation
Quality of life

ABSTRACT

Background: Anecdotal reports suggested that a specialized physiotherapy technique developed in France (the Bleton technique) improved primary cervical dystonia. We evaluated the technique in a randomized trial.
Methods: A parallel-group, single-blind, two-centre randomized trial compared the specialized outpatient physiotherapy programme given by trained physiotherapists up to once a week for 24 weeks with standard physiotherapy advice for neck problems. Randomization was by a central telephone service. The primary outcome was the change in the total Toronto Western Spasmodic Torticollis Rating (TWSTR) scale, measured before any botulinum injections that were due, between baseline and 24 weeks evaluated by a clinician masked to treatment. Analysis was by intention-to-treat.
Results: 110 patients were randomized (55 in each group) with 24 week outcomes available for 84. Most (92%) were receiving botulinum toxin injections. Physiotherapy adherence was good. There was no difference between the groups in the change in TWSTR score over 24 weeks (mean adjusted difference 1.44 [95% CI -3.63, 6.51]) or 52 weeks (mean adjusted difference 2.47 [-2.72, 7.65]) nor in any of the secondary outcome measures (Cervical Dystonia Impact Profile-58, clinician and patient-rated global impression of change, mean botulinum toxin dose). Both groups showed large sustained improvements compared to baseline in the TWSTR, most of which occurred in the first four weeks. There were no major adverse events. Subgroup analysis suggested a centre effect.
Conclusion: There was no statistically or clinically significant benefit from the specialized physiotherapy compared to standard neck physiotherapy advice but further trials are warranted.

1. Background

Cervical dystonia is the commonest type of dystonia with a prevalence of at least six per 100,000 [1,2]. Botulinum toxin injections are the main treatment, which improve neck position and pain [3,4] but often provide inadequate relief [5]. Therefore, alternative treatments are required. Oral medication is usually ineffective [6]. Physiotherapy is used but has a limited evidence-base. Recent systematic reviews of physiotherapy for cervical dystonia [7,8] found only four small (n = 12–40) randomized trials [9–12], testing several combinations of techniques including biofeedback, relaxation therapy, stretching, active exercises and transcutaneous nerve stimulation and concluded no firm recommendations on efficacy could be made.

A well-described, personalized intensive physiotherapy programme (called the Bleton technique after the physiotherapist who developed it) has been used to treat some people with cervical dystonia for several years [13] but has only been evaluated in one small inconclusive trial [12]. We, therefore, did a larger trial to assess whether this technique improved patient outcomes compared to standard physiotherapy advice.

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http://dx.doi.org/10.1016/j.parkreldis.2015.12.010
1353-8020/

2. Methods

2.1. Trial design

This was a two-centre, randomized, parallel-group trial, which compared the Bleton physiotherapy technique with standard neck physiotherapy in adults with primary cervical dystonia. Both groups received standard best treatment, which for most included botulinum toxin injections every three months. The trial was registered with ClinicalTrials.gov (NCT00703287), approved by West Glasgow Ethics Committee and all participants gave written informed consent.

2.2. Trial participants

Eligible adult patients were recruited from two Scottish dystonia clinics provided they had a primary, non-psychogenic, focal cervical dystonia with an abnormal neck position (Toronto Western Spasmodic Torticollis Rating Scale (TWSTR) [14] Part 1A > 0) and were able to give informed consent. Patients being treated with botulinum toxin (type A or B) injections had to be on a stable regimen (same dose and injection pattern over the previous two injections) but those not receiving injections were also eligible providing they were happy to remain off injections for the duration of the trial. Exclusion criteria were secondary cervical dystonia, radicular/myelopathic features where cervical manipulation may be dangerous, fused cervical vertebrae shown on previous x-rays, previous use of the Bleton technique or deep brain stimulation.

2.3. Trial interventions

2.3.1. Individualized specialized physiotherapy regimen

Each centre had an experienced study physiotherapist (4–10 years post-qualification) with broad experience in neurological and musculoskeletal rehabilitation, who was trained for six weeks by Monsieur Bleton in his technique. After initial assessment the physiotherapist developed a personalized regimen for each patient according to the Bleton protocol, which has been described in detail (see appendix and ref [13]). Patients were treated in an outpatient physiotherapy department up to once a week for 45 min per session for 24 weeks.

2.3.2. Standard physiotherapy advice

The control group received standard physiotherapy advice for neck problems (see appendix). Although most patients with cervical dystonia in the UK do not get physiotherapy as standard care, we opted for this as a type of “placebo” to reduce potential bias from simply attending the physiotherapist. Patients were offered sessions every two to four weeks for 24 weeks.

For both groups the physiotherapist recorded attendance, duration and type of treatment per session and frequency of home exercises from their diaries. At the end of the intervention period the physiotherapist rated compliance with the exercise programme as poor, moderate or good, and encouraged patients in both groups to continue with the exercises they had been taught. No other neck physiotherapy was permitted for their dystonia during the one-year trial period. Both groups continued to receive standard care for their dystonia throughout the trial including any botulinum toxin injections according their normal schedule. The dose and site of injections could be adjusted according to clinical need.

2.4. Trial outcomes

The primary outcome was the change from baseline to 24 weeks (end of supervised physiotherapy) in the total TWSTR score. This widely used scale assesses neck position and dystonia-related disability and pain and has a teaching video [14], which the study outcome assessors watched to ensure standardization across the two centres. Secondary outcomes were: (i) change in total TWSTR and its subscores (severity of neck position, disability, pain) at 52 weeks (six months post-treatment to assess whether there was a prolonged benefit especially as patients were encouraged to continue with home exercises after the supervised period); (ii) change in Cervical Dystonia Impact Profile-58 (CDIP-58) score at 24 and 52 weeks, a rigorously developed and validated patient-completed 58 item health impact measure for cervical dystonia, which covers eight dimensions of health (head and neck symptoms, pain, upper limb activities, walking, sleep, annoyance, mood, psychosocial function) [15,16]; (iii) patient and clinician-based global impression of change on a seven point Likert scale (marked/moderate/minimal improvement, no change, minimal/moderate/marked deterioration) at 24 and 52 weeks; (iv) change in a generic quality-of-life measure, the EQ-5D, at 52 weeks; (v) adverse events defined as any unfavourable sign, symptom or illness that developed or worsened during the study, regardless of whether it was considered to be related to study treatment.

2.5. Randomization and masking

Consent and baseline assessments were done by the clinician/nurse involved in the patient's routine care before randomization, who then informed the physiotherapist who randomized each patient by telephoning a central randomization service at the Centre for Healthcare Randomised Trials, University of Aberdeen. Randomization was by a minimization algorithm using centre, the use of botulinum toxin and type of dystonia (predominantly tonic/static head deviation versus marked jerky/tremulous component). Only the treating physiotherapist was informed of the treatment allocation: the clinician/nurse who performed the outcome assessments remained masked throughout. Patients were unmasked but to minimize bias with the self-completed questionnaires the written and verbal information about the trial highlighted that no form of physiotherapy had been shown to be effective in cervical dystonia and that both groups would receive individualized physiotherapy requiring visits between one and four weekly. The Bleton technique was not specifically mentioned. Patients were told not to discuss their physiotherapy with their clinician/nurse.

2.6. Study visits

The baseline visit was performed just before the next botulinum injection in those having injections when their previous botulinum injection would be wearing off. Baseline data collection included demographic data, co-morbidities, current medication, duration of dystonia, details of previous botulinum toxin treatment and baseline assessment of the TWSTR scale, CDIP-58 and EQ-5D. The neck position was recorded on video using a standard protocol. After randomization, the physiotherapist, who was unaware of the baseline scores, arranged their first appointment as soon as possible. Patients were seen by the nurse four, 24 and 52 weeks after the baseline visit to record subsequent botulinum injections (dose and sites), the TWSTR score (with repeat videos), CDIP-58, patient- and nurse-rated global impression of change and any adverse events. In those receiving botulinum injections the four week visit was timed flexibly so that it occurred about four weeks after their last injection (i.e. the time of maximal benefit), whilst the 24 and 52 week visits took place just before their next injection as the toxin was wearing off, matching the baseline visit, to minimize confounding by the botulinum toxin.