



Published in final edited form as:

*J Neurol Neurosurg Psychiatry*. 2014 August ; 85(8): 882–884. doi:10.1136/jnnp-2013-307316.

## Alleviating manoeuvres (sensory tricks) in cervical dystonia

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### Abstract

**Background**—There is limited information on the phenomenology, clinical characteristics and pathophysiology of alleviating manoeuvres (AM), also called ‘sensory tricks’ in cervical dystonia (CD).

**Methods**—Individual data, collected from 10 sites participating in the Dystonia Coalition (<http://clinicaltrials.gov/show/NCT01373424>), included description of localisation and phenomenology of AM collected by systematic review of standardised video examinations. Analyses correlated demographic, neurologic, and psychiatric features of CD patients with or without effective AM.

**Results**—Of 154 people studied, 138 (89.6%) used AM, of which 60 (43.4%) reported partial improvement, 55 (39.8%) marked improvement, and 4 (0.03%) no effect on dystonic posture. Light touch, usually to the lower face or neck, was used by >90%. The presence or location of AM did not correlate with the severity of the dystonia.

**Conclusions**—In this large and comprehensive study of CD, we found no clinical predictors of effective AM. Further studies of sensorimotor integration in dystonia are needed to better understand the pathophysiology of AM.

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**Contributors** NP was involved in the organisation and conceptualisation of the study, gathered the data and wrote the first draft of the manuscript. JH performed the statistical analysis and review of the manuscript. LM was involved in gathering the data, conceptualisation of the study, and review of the manuscript. JJ was involved in the conceptualisation and organisation of the study and review of the manuscript.

**Competing interests** None.

**Ethics approval** Baylor College of Medicine Institutional Review Board.

**Provenance and peer review** Not commissioned; externally peer reviewed.

**Data sharing statement** This data collected through the Dystonia Coalition project is accessible to all the investigators of the Dystonia Coalition. This study was an analysis of data to answer specific questions regarding alleviating manoeuvres which was approved by the Dystonia Coalition steering committee.

Dystonia encompasses a broad range of patterned movements produced by involuntary muscle contractions causing twisting, squeezing and other abnormal postures that are often initiated or worsened by voluntary action, may be associated with overflow muscle activation, and can be categorised by anatomic distribution as either focal, segmental or generalised.<sup>1-4</sup> Frequently, individuals with dystonia adopt a variety of alleviating manoeuvres (AM), also referred to as sensory tricks, or the geste antagoniste, to correct the dystonic posture or stop the abnormal movement.<sup>5-7</sup> We believe that AM is the more appropriate term for this phenomenon than the classic term 'sensory trick' because the latter suggests that only sensory input is required, and the word 'trick' implies that it is 'fake'.<sup>6</sup> Furthermore, the term 'sensory trick' has been wrongly interpreted as implying psychological origin of dystonia.

The presence of AM supports the emerging understanding that dystonia as a disorder of sensorimotor integration.<sup>6-9</sup> Only a few studies describe the phenomenology of AM, primarily in cervical dystonia (CD) and blepharospasm.<sup>10-15</sup> These descriptions, however, are often inadequate or are based on small cohorts. To overcome these limitations, we systematically collected demographic and clinical data on CD patients with and without AM, in a large multicentre cohort of patients enrolled in Dystonia Coalition Project 2.

## METHODS

This study was approved by the Baylor College of Medicine Institutional Review Board and the Dystonia Coalition steering committee. We analysed all the data collected from 164 CD patients enrolled across 10 sites in Project 2 of the Dystonia Coalition (<http://clinicaltrials.gov/show/NCT01373424>). All patients were examined and videotaped according to a standard protocol (3 months after last botulinum toxin injection), which included instructions to patients to demonstrate the most effective 'sensory trick'. Descriptive details of AM were collected for locations of effective AMs, laterality of touch (eg, ipsilateral hand to face), type of AM and associated degree of improvement through standardised video examinations. Some centres captured several manoeuvres, while others demonstrated the single best manoeuvre. The touch AMs were classified by a single video rater (NP) as either light touch (LT) or forcible touch (FT), based on a published rating method.<sup>7, 12</sup> The effectiveness of the AM was subjectively determined by the rater as marked (> 75%) improvement, partial improvement, or no effect (< 25% improvement). Those CD patients with effective AM, defined as partial or marked improvement of their CD, were compared to patients without effective AM with respect to demographic, neurological and psychiatric diagnoses, as well as with the scores on Global Dystonia Rating Scale (GDRS) and Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS).

Statistical analyses included t tests for quantitative outcomes or exact Pearson  $\chi^2$  tests for nominal outcomes. All tests were two-sided. We also conducted multiple linear regression analyses to adjust for possible confounding effects. p Values less than 0.05 were considered significant.

## RESULTS

We identified 164 participants, 30 of whom were enrolled from the Baylor College of Medicine Movement Disorders Clinic. The mean age was  $59.5 \pm 10.9$  years, and mean duration of dystonia was  $15.3 \pm 11.5$  years (table 1). Ten patients were excluded due to insufficient video data; therefore, 154 CD patients were available for analysis. Of the 138 (89.6%) that reported the use of AM, 55 (39.8%) experienced marked improvement, 60 (43.4%) partial improvement, and 4 (0.03%) no effect; the degree of effectiveness (partial or complete) was not available for analysis in 19 subjects. Of the patients using AM, 125 (90.5%) used LT and 13 (9.4%) used FT. The following AMs were used: 77 subjects touched their lower face, 61 chin, 46 neck, 16 upper face and 2 shoulder, using either their ipsilateral, contralateral or both hands to stabilise the head position (table 2). We were unable to correlate a location of AM to direction of dystonic posture due to the variable combination of dystonic postures in each patient.

Patients with AMs (n=138) had significantly higher unadjusted GDRS total scores compared to the 16 patients who did not use AM ( $p=0.05$ ). However, after adjusting for age, duration of dystonia, and presence of psychiatric conditions using multiple linear regression analysis, the difference in GDRS scores and TWSTRS total scores between the two groups was not significant ( $p=0.13$  and  $p=0.37$ , respectively). We also conducted multiple linear regression analyses confined to the 138 patients in the AM group, the locations of AM (upper face, lower face, chin, and posterior neck) as explanatory variables in addition to age, duration of dystonia, and presence of psychiatric conditions (depression and/or anxiety). The results indicated that the location of AM did not correlate with either the GDRS total score ( $p=0.82$ ) or the TWSTRS total score ( $p=0.85$ ).

## DISCUSSION

To our knowledge, this is the largest cohort of patients in whom the characteristics of AMs were systematically determined through standardised clinical evaluations and videos. Age and gender distribution is similar to other reports.<sup>16</sup> We found that 83% of CD patients noted partial or marked benefit with use of AM which is similar to other published reports.<sup>10, 12–14</sup> One study, involving 32 patients with focal or segmental dystonia, found that 90.5% of patients reported improvement with AM that was later confirmed by clinical examination.<sup>13</sup> The locations and characteristics of the most common AMs, lower face, chin and neck, reported in our study is similar to those previously described.<sup>12, 13</sup> In a questionnaire-based study of 33 CD patients, 8 self-reported marked relief of CD with application of AMs with 25 reporting partial improvement of abnormal posture.<sup>10</sup> In this study, 42% of the patients used ‘forcible trick’, whereas only 9.4% of our patients required FT. We also found that CD patients primarily use LT, but occasionally have to use FT, particularly with the hand ipsilateral to the torticollis or laterocollis.<sup>6</sup> Attenuation of muscle activity has been demonstrated with the application of LT in a specific area of the body (typically related to the area of dystonia) with electromyography (EMG).<sup>14, 17, 18</sup>

The mechanism by which AM improves dystonia is not well understood. Although ‘counter pressure’ has been suggested as a mechanism for AM in patients with CD, careful

observation reveals that LT, rather than FT, to the chin ipsilateral to the direction of head rotation is typically sufficient to bring the head into primary position. A two-phase model using EMG in CD patient was proposed. In this model, abnormal head posture is first normalised by counter pressure of volitional antagonistic muscle activity after which the position is stabilised by sensory input.<sup>17</sup> Furthermore, shorter duration of illness, and minimal impairment of visuotactile discrimination, were reported in those patients with nearly complete resolution of CD with use of AMs compared to those with partial or no relief.<sup>19</sup> Although patients and clinicians often make the observation that AMs are more common early and become less effective over time, our study found no correlation between duration or severity of dystonia and the efficacy of AM. It is possible that the long duration and relatively marked severity of dystonia in our patients (table 1) accounts for the lack of correlation in our cohort of patients. Besides touching the ipsilateral face, there are many other AMs that have been reported in the literature that include voluntary abduction of the ipsilateral shoulder, touching the cheek, chin or other areas of the head and face, resting the occiput against the wall when sitting or standing, placing objects (eg, books) on top of the head or simply holding one's hand above the vertex of the head without even touching the head, bending the trunk forward or yawning.<sup>6, 12, 14</sup> Most patients note that they, rather than someone else, must do the touching, although in some cases a mere imagination of AM can improve the abnormal posture or movement.<sup>7, 20</sup> Proprioceptive vibration-induced illusion of movement of an artificial hand, using the rubber hand paradigm to test sensory ownership of limb, suggests that patients with focal dystonia have deficits in visual-tactile-proprioceptive integration involving the inferior parietal cortex and the cerebellum.<sup>21, 22</sup> The broad spectrum of AMs used by patients with CD and other forms of dystonia strongly suggests that a variety of sensorimotor processes are involved in mediating this phenomenon. Several studies have demonstrated abnormal kinaesthetic perception,<sup>23</sup> and impaired integration of proprioceptive input<sup>24, 25</sup> in the pathophysiology of CD, indicating that the AM may augment peripheral sensory inputs which result in improved posture. Using EMG and positron emission tomography in patients with CD, reduced activation of the supplementary motor and primary sensorimotor cortex contralateral to the side of dystonic posture, was found with the application of AM.<sup>26</sup> This finding of hyperactive parietal cortex during the execution of AM is consistent with other studies showing that this area, normally involved in multimodal sensory integration, plays a key role in the effectiveness of AM.<sup>19</sup>

Limitations to this study include the retrospective review of the demographic and clinical data based on patient recollection through the Dystonia Coalition study, and the variability in videotaped descriptions and demonstrations of the AMs, with the camera in a fixed position perpendicular to the face. The characterisations of AM as partial versus marked, and LT versus FT, were determined by subjective interpretation of the video examination. Finally, the relatively small number of patients in this cohort who did not use AM (n=16) relative to those who did (n=138) limited the statistical analyses. Nevertheless, this study provides the largest and most comprehensive description of AMs in patients with CD. Phenomenological characterisation of AMs and their effect on pain can be applied to therapeutic strategies. We and others, for example, have used the information about AMs to design specific cervical braces and other appliances that patients use to facilitate their unique AMs.

## Acknowledgments

**Funding** Subject recruitment and assessment was made possible through the Dystonia Coalition from the Office of Rare Diseases Research at the National Center for Advancing Translational Sciences (NIH grant NS 065701) and the Dystonia Medical Research Foundation.

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**Table 1**

Demographics, effectiveness and symptoms severity in patients with effective alleviating manoeuvres who used forceful touch or light touch only (n=138)

	Used forceful touch at any location (n=13)	Used light touch only at any location (n=125)	Test of difference p value
Age (years)	62.9±10.5 (46–83)	59.5±10.6 (29–80)	0.34
Duration of dystonia (years)	18.0±13.0 (0–39)	15.1±11.3 (0–60)	0.44
GDRS (total score)	13.6±10.1 (3–34)	8.6±5.1 (1–37)	0.15
TWSTRS (total score)	17.2±6.8 (7–29)	16.2±5.6 (1–27)	0.58
Gender (%)			0.73
Female	11 (85)	94 (75)	
Male	2 (15)	31 (25)	
Effectiveness of AM (%)			0.28
Partial	8 (73)	52 (48)	
Complete	3 (27)	52 (48)	
No effect	0	4 (4)	

Partial missing information of the 138 who reported effective alleviating manoeuvres: age, 9 patients; duration of dystonia, 9 patients; GDRS, 11 patients; degree of improvement, 19 patients.

AM, alleviating manoeuvres; GDRS, Global Dystonia Rating Scale; TWSTRS, Toronto Western Spasmodic Torticollis Rating Scale.

**Table 2**

Locations and characteristics of alleviating manoeuvres

	Upper face (n=16)	Lower face (n=77)	Chin (n=61)	Posterior neck (n=46)	Shoulder (n=2)
Ipsilateral, LT	13	59	48	29	1
Ipsilateral, FT	1	4	4	3	0
Ipsilateral, URT	2	4	2	5	0
Contralateral, LT	0	6	3	2	0
Contralateral, FT	0	0	0	0	0
Contralateral, URT	0	0	0	0	0
Bilateral, LT	0	4	4	4	0
Bilateral, FT	0	0	0	3	1
Bilateral, URT	0	0	0	0	0

Data collected through systematic review of videotaped neurological examinations. Some patients demonstrated effective tricks in multiple locations.

bilateral, 2 hands touching body; contralateral, hand crossing midline to touch opposite side of the body; FT, forceful touch, a forceful pressure applied to improve dystonic posture; ipsilateral, hand touching same side of body; LT, Light touch, a gentle touch to improve dystonic posture; URT, unrated touch.