Alleviating Maneuvers (Sensory Tricks) in Cervical Dystonia

Neepa Patel, MD[1]; John Hanfelt, PhD[2]; Laura Marsh, MD[3]; Joseph Jankovic, MD[1] for the Dystonia Coalition

[1]Parkinson's Disease Center and Movement Disorders Clinic, Department of Neurology; Baylor College of Medicine, Houston, Texas
[2]Department of Statistics and Bioinformatics; Emory University, Atlanta, Georgia
[3]Department of Psychiatry and Neurology; Baylor College of Medicine, Houston, Texas

This consortium (Dystonia Coalition) is a part of NIH Rare Diseases Clinical Research Network (RDCRN). Funding has been provided by 1U54NS065701-01.

BACKGROUND

- Dystonia encompasses a broad range of movements defined as sustained, patterned involuntary muscle contractions causing twisting and abnormal posture[1-3].
- The sensory trick (ST), otherwise known as the *geste antagnoiste* is a classic feature of most focal dystonias.
- This maneuver has traditionally been used to describe a light touch to an area of the body which improves the abnormal posture.
- There are few studies describing the phenomenology of sensory tricks primarily in cervical dystonia (CD) and blepharospasm[4-8].
- However these studies were performed in a single center describing a small number of patients.

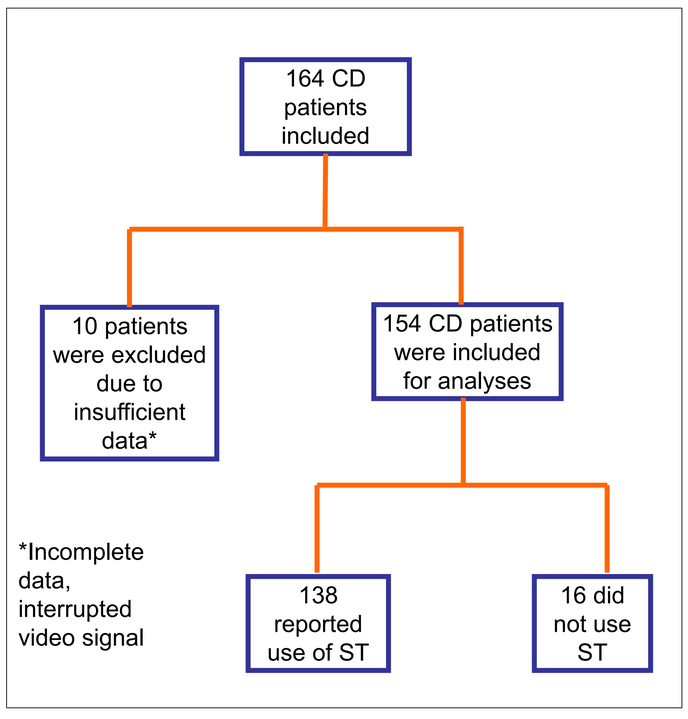
OBJECTIVES

- To determine the demographic and clinical differences between patients with and without ST, in a large multicenter cohort of patients with cervical dystonia (CD) enrolled in the Dystonia Coalition registry

METHODS

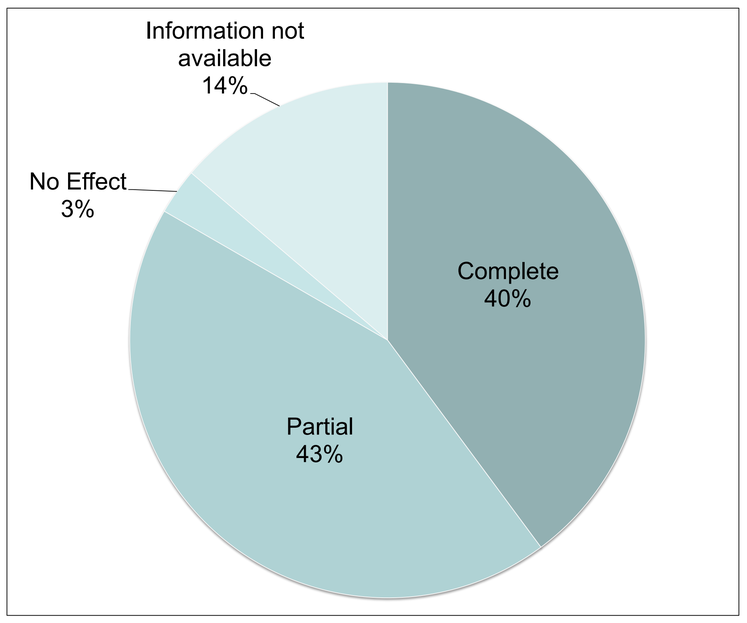
- This is an institutional review board approved retrospective study
- We analyzed the data collected from 164 cervical dystonia patients enrolled in 9 sites in the Project 2 arm of the Dystonia Coalition project (http://clinicaltrials.gov/show/NCT01373424) by November 2012.
- CD with effective ST, defined as partial or complete improvement of the abnormal posture, were compared to CD patients without effective ST on demographic and neurologic features and psychiatric diagnoses.
- Details regarding localization and phenomenology of effective ST and degree of improvement were collected initial data collection:
  - Demographic variables
  - Global Dystonia Rating Scale (GDRS)
  - Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS)
- Systematic review of standardized video examinations for descriptive details of ST:
  - Site and characteristics of ST
  - Degree effectiveness
- Analyses used t-test or exact Pearson chi-square tests (for nominal outcomes).

Figure 1: Inclusion Criteria



RESULTS

Figure 2: Effects of Sensory Trick on CD (n=138)



Data was collected through systematic review of video examination by a single examiner. Results may vary from the original data captured at enrollment of the subject.

Table 1: Demographics and Symptom Severity

	Used ST (n = 138)	Did Not Use ST (n = 16)	Test of Difference
Age (years)	59.8 ± 10.6 (29-83)	59.7 ± 10.5 (43-77)	P = 0.98
Duration of dystonia (years)	15.3 ± 11.4 (0-60)	11.3 ± 7.5 (2-27)	P = 0.08*
GDRS (total score)	9.0 ± 5.7 (1-37)	5.9 ± 4.2 (0-13)	P = 0.05**
TWSTRS (total score)	16.3 ± 5.7 (1-29)	13.8 ± 5.9 (4-23)	P = 0.11*
Psychiatric Conditions:			P = 0.42
Present	48 (37%)	4 (25%)	
Absent	83 (63%)	12 (75%)	

** statistically significant for unadjusted variables.
* Trend towards significance for unadjusted variables

Table 2: Locations and Characteristics of ST

	Upper Face (n = 16)	Lower Face (n = 77)	Chin (n=61)	Neck (n = 46)	Shoulder (n = 2)
Ipsilateral light touch	13	59	48	29	1
Ipsilateral forceful touch	1	4	4	3	0
Ipsilateral unrated touch	2	4	2	5	0
Contralateral light touch	0	6	3	2	0
Contralateral forceful touch	0	0	0	0	0
Contralateral unrated touch	0	0	0	0	0
Bilateral light touch	0	4	4	4	0
Bilateral forceful touch	0	0	0	3	1
Bilateral unrated touch	0	0	0	0	0

- Patients with ST had significantly higher GDRS total scores compared to patients who did not use ST (p=0.05)
- However, after adjustment for age, duration of dystonia and presence of psychiatric conditions using multiple linear regression analysis comparing the ST vs. non-ST group.
  - There was no significant difference in GDRS scores (p= 0.13)
  - There was no significant difference in TWSTRS total scores (p= 0.37).

DISCUSSION

- To our knowledge this is the largest cohort of patients describing the characteristics of STs that were systematically determined through clinical examination.
- Limitations to this study include:
  - Retrospective nature of data review
  - Variability in videotaped demonstrations of ST between centers.
- The presence of improvement with "forceful touch" demonstrates that the ST benefits may also be present with motor input.
- Our findings are similar to others in that the benefits of ST may be earlier in the course of the disease[9].
- Abnormalities in proprioceptive, spatial and temporal sensory discrimination have been identified[10-11].
- In patients with CD a two-phase model in which abnormal head posture is first normalized by counter pressure or volitional antagonistic muscle activity after which the position is stabilized by sensory input[12].
- The presence of the ST supports emerging theories that dystonia is a disorder of sensorimotor integration[13-14].

CONCLUSION

- This study demonstrates that the ST does not involve just "sensory" input and that it is effective rather than "fake" as implied by the word "trick".
- We propose that a more appropriate term for this phenomenon is "alleviating maneuver" (AM) which could be either motor or sensory in nature.
- Future studies should be directed towards:
  - Further clarification of the pathophysiological mechanism of the AM
  - Therapeutic strategies that utilize the benefits of the AM

REFERENCES

1. Colosimo, C., Berardelli, A., 2011. Clinical phenomenology of dystonia. Int Rev Neurobiol. 98, 509-24.
2. LeDoux, M.S., 2012. Dystonia: phenomenology. Parkinsonism Relat Disord. 18 Suppl 1, S162-4.
3. Phukan, J., Albanese, A., Gasser, T., Warner, T., 2011. Primary dystonia and dystonia-plus syndromes: clinical characteristics, diagnosis, and pathogenesis. Lancet Neurol. 10, 1074-85.
4. Jahanshahi, M., 2000. Factors that ameliorate or aggravate spasmodic torticollis. J Neurol Neurosurg Psychiatry. 68, 227-9.
5. Jankovic, J., Leder, S., Warner, D., Schwartz, K., 1991. Cervical dystonia: clinical findings and associated movement disorders. Neurology. 41, 1088-91.
6. Ochudlo, S., Drzyzga, K., Drzyzga, L.R., Opala, G., 2007. Various patterns of gestes antagonistes in cervical dystonia. Parkinsonism Relat Disord. 13, 417-20.
7. Martino, D., Liuzzi, D., Macerollo, A., Aniello, M.S., Livrea, P., Defazio, G., 2010. The phenomenology of the geste antagoniste in primary blepharospasm and cervical dystonia. Mov Disord. 25, 407-12.
8. Muller, J., Wissel, J., Masuhr, F., Ebersbach, G., Wenning, G.K., Poewe, W., 2001. Clinical characteristics of the geste antagoniste in cervical dystonia. J Neurol. 248, 478-82.
9. Kagi, G., et al., 2013. Sensory tricks in primary cervical dystonia depend on visuotactile temporal discrimination. Mov Disord.
10. Tinazzi, M., Fiorio, M., Fiaschi, A., Rothwell, J.C., Bhatia, K.P., 2009. Sensory functions in dystonia: insights from behavioral studies. Mov Disord. 24, 1427-36.
11. Bradley, D., et al., 2010. Comparing endophenotypes in adult-onset primary torsion dystonia. Mov Disord. 25, 84-90.
12. Schramm, A., Reiners, K., Naumann, M., 2004. Complex mechanisms of sensory tricks in cervical dystonia. Mov Disord. 19, 452-8.
13. Abbruzzese, G., Berardelli, A., 2003. Sensorimotor integration in movement disorders. Mov Disord. 18, 231-40.
14. Hallett, M., 2009. Dystonia: a sensory and motor disorder of short latency inhibition. Ann Neurol. 66, 125-7.