Clinical Similarities in Cervical Dystonia Patients With and Without a History of Painful Trauma

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Why dystonia, generalized or focal, occurs is unknown. Increasing evidence suggests that generalized dystonia has a strong genetic component. Further, that in families with generalized dystonia, the rate of focal dystonia in other family members is increased.[1] My own observations of the reverse; that is, families with focal dystonia having members with generalized dystonia have failed to yield such a reverse association, at least by history.[2] With respect to focal dystonia, Waddy, et al[3] observed a high frequency of focal dystonic signs in family members of patients with focal dystonia, reinforcing a genetic etiology for focal dystonia.

Among the alternate factors which have been speculated to at least initiate dystonic symptoms is trauma. A specific characteristic of the trauma felt critical to the precipitation of symptoms is pain, and that peripheral pain somehow alters central mechanisms leading to central overactivity and consequent peripheral muscular overcontraction, presumably dystonic contraction.[4,5,6]

Sheehy and Marsden,[7] and more recently, Jankovic and Van Der Linden,[8] have speculated on antecedent trauma as a precipitating factor in the production of focal cervical dystonia. In the present study, cervical dystonia patients who described painful injury to the head, neck or shoulder one year or less prior to onset were designated as “history of trauma.” None of these patients recalled neurologic deficits such as loss of consciousness, paresthesia or paresis. Data were collected per protocol regarding personal family, neurological and general medical history, laboratory investigation and a protocol neurologic examination performed by this author. The proposition is that if trauma is an independent, pathogenetic factor in dystonia, one or more characteristics may differ between the two populations.

Between January, 1988 and September, 1991, 198 consecutive patients with cervical dystonia were evaluated. 41, or 21%, described a painful injury to the head, neck or shoulder within one year of the recalled onset of cervical dystonic symptoms. This contrasts with a figure of 11% given by Jankovic and associates from a recently reported large series.[9] The gender ratio between the two groups is similar, with approximately 3/4ths of the patients female in both groups. The age of onset, standard deviation and range, was similar for the two groups in the mid 40s. Duration of symptoms at the time of evaluation was comparable as well at about 10 years.

Among the patient characteristics contrasted was the frequency of light eye color, defined as gray, blue, green or hazel. Although a controversial issue debated in the past between Korein[10] and Lang et al,[11] my own observations from a large pool of patients personally evaluated while still at Mayo Clinic Rochester[12] revealed a high frequency of light eyes in the torticollis population which was statistically significantly different from a control population of routine neurologic patients. We have repeated that observation with the same finding in patients evaluated in the last four years in Arizona contrasted with a comparable population of patients with cervical spondylosis. At any rate, on the basis of eye color, these two groups these with and without trauma are indistinguishable.

Analysis of the initial symptom in the two populations suggests no major differences. Although pain was more commonly reported by those with a history of trauma, it was also a selection criterion.

Next, we analyzed the features of the dystonia itself. In each group, about 1/3 demonstrated dystonic symptoms outside of the neck similar to the report of Jankovic and associates[9] and our own report last year at the ANA in Seattle.[13] Truong and associates[14] have pointed out that traumatic torticollis is apt to be tonic in presentation. Overall, however, the frequency of phasic versus tonic movements in these two populations was similar. An equal, slight preponderance of chin rotation toward the patient's left was noted in both groups. An identical frequency of accelerometer measured head and hand tremor was noted. Prevalence of tremor in this population is comparable to that described by Jankovic and associates.[9]

Analysis of the distribution of extranuchal sites for dystonia, in addition to similar overall prevalence, shows relative similarity with the possible exception of brachial and oral-periocular sites, the former more prevalent, the latter less prevalent when there was a history of injury.

Others and I have previously reported on the high frequency of occurrence of recalled family history of essential tremor and other dystonic symptoms among those with cervical dystonia.[2,9] This population is similar to those previously reported, with the most common recalled family history that of an essential type of tremor followed by both essential tremor and focal dystonia in the family, followed by scoliosis and about an equal frequency of spasmodic torticollis or facial movement disorder.

As we previously reported, family history of Parkinson's disease is not more frequent among those with torticollis when contrasted with a control population,[2] and no different whether or not there is a history of trauma in this investigation. Thus, it is not obvious that a family history of a movement disorder increases the risk of dystonia following on the heels of injury.

We have previously shown that although thyroid dysfunction is common among, especially, females with torticollis, it is only weakly statistically different from a control population of similar aged women with cervical spondylosis.[2] History of bona fide thyroid dysfunction or measured abnormalities in serum thyroxin or serum thyroid stimulating hormone were comparable in these two populations. We also had shown last year that at this meeting that elevated antinuclear antibody titers are a bit more frequent in females with spasmodic torticollis versus a control population. However, the frequency of elevated antibody titers to thyroid tissue, antinuclear antibody or rheumatoid factor is similar irrespective of history of injury.

At the World Congress of Movement Disorders convening in Munich this June, we will be reporting on an analysis of psychiatric features comparing cervical dystonia patients with patients with chronic spine pain. That study shows a high frequency of antecedent psychiatric diagnosis in both those with cervical dystonia and pain control patients, but an increased risk of depression in the course of dystonic illness, including a higher risk for a psychiatric referral and diagnosis in those with cervical dystonia.[15] Comparing in this study those with and without a history of trauma, antecedent and postcedent psychiatric diagnosis is similar, and the frequency of documented anxiety by the Purdue Anxiety Index from the MMPI, or depression from the depression scale of the MMPI, or both, is similar in the two populations.

Since the submission of this abstract, we have completed the data analysis of other features of these patients, including the results of MRI findings in this and a larger population of 212 patients which will be reported at the Movement Disorder Society meeting in Toronto this October.[16] 27% of those without a history of trauma and 32% of those with a history of trauma has undergone MRI investigation of the head. That post priori study showed a surprisingly high frequency of abnormality, and in our CD population, nearly 43%. There was a slightly higher probability of an abnormal MRI, meaning unusual ventricular dilation well beyond normal for age, and focal much more than multifocal white matter changes also judged unusual for age. Gordon and associates previously had suggested that anomalous ventricular configuration by CT scan might be associated with traumatically induced dystonia.[17]

Furthermore, a comparison of the rate of perinatal stressors, peripubertal symptoms, such as evidence of scoliosis, transient tremor, or transient writer's cramp as well as left-handedness, was more common in those with a history of trauma than those without. These last findings are similar to those observed by Jankovic and Van Der Linden[8] and raise the possibility that among the predisposing factors to a dystonic consequence to injury may be these alternate, perhaps epigenetic, central nervous system events.

The frequency of exposure to amphetamine or neuroleptics antecedent to the onset of dystonia in these two populations was similar. The same is true of prior infection. Furthermore, the probability of remission was comparable in the two populations. Finally, there appeared to be no addictive risk with presence of both family history of movement disorder and perinatal stress.

In the main, these two populations, those with and without a history of trauma, are very comparable, suggesting that with respect to cervical dystonia, similar biologic factors are at play in the dystonic consequence to painful trauma. This, in my judgment, is a different group of patients than those who have painful injury with focal spasm which may distort the posture of the neck or shoulder. Such tonic muscle contraction is not identical to that observed in otherwise "idiopathic" focal cervical dystonia. Thus, the designation "cervical dystonia" should be reserved for unequivocal dystonic spasm, while the term "torticollis" remains a generic term referring to a distortion of head and neck posture, the etiology for which might be different and more heterogenous than that of dystonia.

We are currently involved in an analysis of multichannel EMG recordings in those whom we think have post traumatic focal spasm versus those with more typical dystonia.

We conclude, therefore, there are no major differences between cervical dystonia patients with or without a history of antecedent painful injury. A possible exception may be a slightly increased risk of antecedent perinatal stress, transient peripubertal dystonic symptoms, left-handedness, MRI abnormalities, or cervical brachial dystonia in those with injury. Consequently, trauma may either unmask dystonic diathesis already biologically present, or represent a coincidental occurrence. These observations do not exclude traumatic origin of other forms of focal muscle spasm which may distort spine posture.

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