Neurology 2005 Mar;64(Suppl 1):A200 ABS-P03.145
2005040515

P03.145
Botulinum toxin treatment for oromandibular dystonia and bruxism in patients with Tourette syndrome

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OBJECTIVE: To draw attention to oromandibular dystonia (OMD), including bruxism, relatively high coexistence of dystonia in patients with Tourette syndrome (TS) and its treatment with botulinum toxin (BTX).
BACKGROUND: OMD typically occurs as parts of primary, adult-onset, cranial dystonia, but it can also occur after local trauma or surgery, as a consequence of exposure to dopamine receptor blocking drugs (tardive dystonia), and in association with other disorders including TS. Some patients with OMD also have bruxism, often misdiagnosed as temporomandibular joint (TMJ) syndrome. While the exact prevalence of OMD and bruxism in TS is unknown, we have evaluated several patients with TS in whom this form of focal dystonia was among the most troublesome symptoms, requiring BTX treatment.
DESIGN/METHODS: Patients diagnosed with TS according to the Tourette Syndrome Classification Study Group criteria, evaluated in our Movement Disorders Clinic and also diagnosed with OMD and/or bruxism and treated with BTX were selected for this study. Besides diagnosis of TS, in order for patients to be included in this study their symptoms must have been of sufficient severity to interfere with chewing or speech, despite medical treatments, and at least one follow up visit after treatment with BTX. Response to treatment, previously published outcome measure, was rated on a 0 to 4 clinical rating scale in which 4 is equal to total abolishment of dystonia.
RESULTS: Of the 107 patients with TS and dystonia in our database, 17 were diagnosed with OMD and/or bruxism and 10 (6 male) patients required BTX treatment. The mean age at initiation of BTX treatment was 18.3 ± 4.1 years (range: 13.4 - 65.3), the mean duration of TS symptoms 36.9 ± 16.4 years (range: 5 - 58.9), and OMD/bruxism symptoms 19.7 ± 10.0 years (range: 2.7 - 44.2). The mean duration of BTX treatment was 3.0 ± 1.9 years (range: 0.3 - 9.7) administered in a total of 71 injection visits. The mean dose of BTX was 63.8 ± 12.3 mouse units (MU) (30 - 150), per side for the masseter muscles, 37.5 ± 12.5 MU (25 - 50) per side for the lateral pterygoids, and 43.6 ± 10.7 MU (10 - 100) for the submental muscle complex. The mean total duration of response was 14.4 ± 12.0 weeks (12 - 50), latency to onset of benefits was 4.6 days (1 - 21). The mean peak effect on a scale of 0 to 4, in which 4 was equal to total abolishment of dystonia, was 3.15 ± 0.9. Of these 10 patients, 5 had intense premonitory sensory symptoms; 4 derived significant relief of these symptoms from BTX. Two subjects reported having experienced dysphagia in at least one visit and one subject reported having experienced dysarthria with BTX. Complications such as dysphagia and dysarthria were reported in 5 of all treatment visits (7%).

CONCLUSIONS: The high prevalence rate of dystonia (7.3% of all patients) in patients with TS suggests that some patients with TS may have an increased risk for dystonia. BTX administered by skilled practitioners is a safe and effective treatment in OMD/Bruxism associated with TS.

Disclosure: Dr. Silay has nothing to disclose. Dr. Jankovic has received personal compensation for activities with: Allergan, Boehringer Ingenheim, Dimensional Healthcare, Healthworld, Pfizer, Prestwick, Valeant. Dr. Jankovic has received personal compensation in an editorial capacity for Medlink, NICP. Dr. Jankovic has received financial support for UCB Pharma: Levatiracetam in ET; Boehringer Ingelheim: NS 2330 in early PD; Guilford Pharmaceuticals: Phase 2 study of GPI 1485 in PD; Prestwick: Tetrabenazine Withdrawal Study in HD; Prestwick: Placebo Controlled Trial of Tetrabenazine.

American Academy of Neurology (AAN) 57th Annual Meeting, April 9 - 16, 2005, Miami Beach, FL