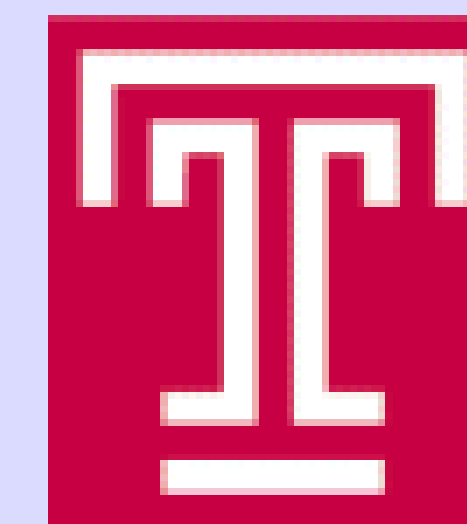


Botulinum Toxin Type A Treatment For Traumatic Brain Injury-Induced Jaw-Opening Oromandibular Dystonia

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ABSTRACT

Setting: Tertiary care rehabilitation center and electrodiagnostic laboratory **Patient:** 18-year-old female with severe traumatic brain injury. **Case:** 18-year-old female with severe traumatic brain injury due to a motor vehicle collision who sustained a right subdural hematoma and diffuse axonal injury along with right orbital and mandible fractures and remained minimally responsive at admission. She had upper motor neuron syndrome, severe spasticity, and hyperreflexia with minimal voluntary control. The patient also presented with severe jaw-closing impairment, which affected her oral hygiene, swallowing mobility, and speech presentation. Electrodiagnostic studies demonstrated hyperactivity of mylohyoid and anterior digastric muscles with only few units in temporalis and masseter muscles. Under electromyographic guidance, the patient underwent trial of marcamine to the dystonic muscles, leading to a 90% closure of the mandible passively; however, her jaw remained open due to contracture of the connective tissues and weakness of the jaw closing muscles. Subsequently, the patient returned for botulinum toxin A injection to bilateral mylohyoid two weeks after the marcamine trial. Ten units of botulinum toxin type A was injected to the mylohyoid bilaterally with electromyographic guidance. The patient was then introduced to oromandibular rehabilitation, passive positional assistance using a helmet with chinstrap to support the jaw and stretching of the connective tissues. **Result:** There was marked improvement of oromandibular muscle control and she was able to participate in speech therapy. At two-month follow-up the patient had salivary control, improved oral care and facial appearance, and was beginning to communicate in simple words. **Discussion:** This is the first reported traumatic brain injury case, to our knowledge, of effective botulinum toxin A treatment in a patient with jaw-opening oromandibular dystonia in the literature. **Conclusion:** Accurate diagnosis and chemodenervation treatment for unopposed jaw-opening can be useful to ensure improvement of oromandibular dynamics.

CASE DESCRIPTION

A healthy 18-year-old female unrestrained driver was in a motor vehicle collision and sustained a right subdural hematoma, diffuse axonal injury, and right orbital, mandibular fractures. She underwent open reduction internal fixation of her mandible and craniectomy and subsequent cranioplasty months later. Her hospital course was complicated with ventilation dependent respiratory failure, seizures, shunt placement, infections and sympathetic storm. She subsequently was transferred to inpatient acute rehabilitation, subacute nursing facility and again to inpatient rehabilitation five months post-injury. Physical examination demonstrated bilateral hemiparesis, severe spasticity, hyperreflexia with minimal voluntary control. The patient also revealed severe jaw-closing impairment, which affected her oral hygiene, swallowing and speech that impeded the nutrition and progress in rehabilitation. Her jaw-closing impairment was assumed to be due to her mandibular fractures prior to her second rehabilitation admission. However, a second maxillofacial surgery evaluation suggested that her jaw-closing impairment was secondary to hyperactivity of the hyoid muscles and not due to temporomandibular joint pathology or mandibular fractures. Electrodiagnostic study demonstrated hyperactivity of mylohyoid and anterior digastric muscles with very few to no voluntary motor units in temporalis and masseter muscles. A treatment plan with a trial of marcamine injection followed by botulinum toxin A injection in conjunction with oromandibular rehabilitation (passive positional assistance using a helmet with chinstrap to support the jaw and stretching of the connective tissues) was developed. She made remarkable improvement in speech, swallowing and function after the brain injury rehabilitation and was discharged home.

EMG Summary Table	Spontaneous					MUAP			Recruitment Pattern
	IA	Fib	PSW	Fasc	H.F.	Amp	Dur.	PPP	
L. Anterior Digastric CN 5, V3	N	None	None	None	None	N	N	N	Discrete
L. Mylohyoid CN 5, V3	N	None	None	None	None	N	N	N	Mild Red
L. Tongue CN 12	N	None	None	None	None	N	N	N	Mod Red
L. Pterygoid CN 5, V3	N	None	None	None	None	N	N	N	Discrete
L. Temporalis CN 5, V3	N	None	None	None	None				No Activity
L. Masseter CN 5, V3	N	None	None	None	None				No Activity
R. Anterior Digastric Cn 5, V3	N	None	None	None	None	N	N	N	Mod Red
R. Mylohyoid CN 5, V3	N	None	None	None	None	N	N	N	Mild Red
R. Tongue CN 12	N	None	None	None	None	N	N	N	Mod Red
R. Pterygoid CN 5, V3	N	None	None	None	None	N	N	N	Mod Red
R. Masseter CN 5, V3	N	None	None	None	None	N	N	N	Discrete

Table 1. Electromyographic results demonstrates increased activation of the mylohyoid, anterior digastric and poor activation of temporalis and masseter muscles.

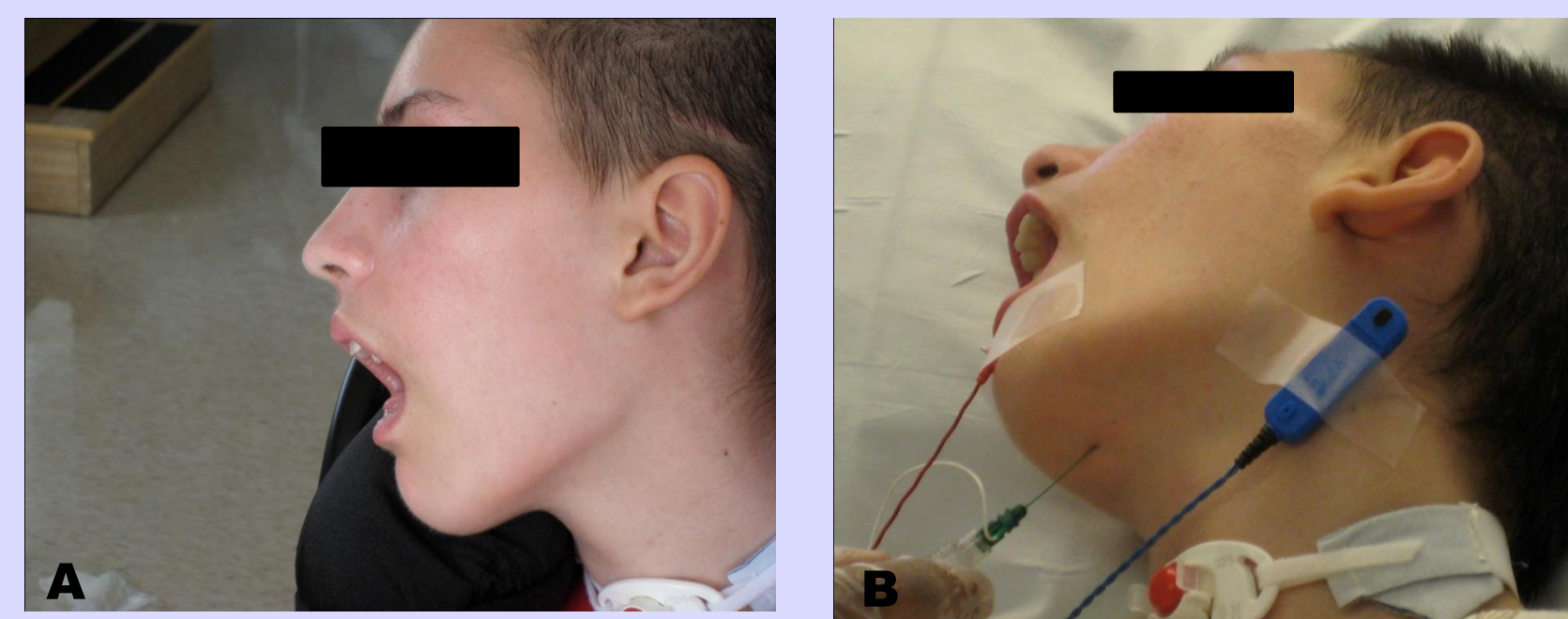


Figure 1. (A) The patient with jaw-opening features (B) Under needle electromyographic guidance, the patient underwent botulinum toxin A injection to the mylohyoid muscle.



Figure 2. (A) Passive positional assistance using a helmet with chinstrap to support the jaw (B) Two months post-botulinum toxin treatment shows jaw closure and improved facial appearance.

DISCUSSION

Chemodenervation with the use of botulinum toxin has emerged as an effective treatment of symptomatic abnormalities in dystonia with oromandibular symptoms within the past few years. To our knowledge, this case study is the first reported traumatic brain injury case of effective botulinum toxin type A treatment in a patient with jaw-opening oromandibular dystonia in the literature. In this case study, our patient had hyperactive mylohyoid and digastric muscles causing unopposed jaw-opening oromandibular dystonia secondary to her traumatic brain injury and mandibular fracture. Due to her prolonged hospital course, she developed soft tissue contractures of her mylohyoid and digastric muscles in addition to hyperactivity and also contracture of the TMJ. After a trial of chemodenervation with marcamine she was able to achieve approximately 90% closure of the mandible passively by the examiner. However, the contracture of the connective tissues caused incomplete closure of her mouth. At rest she remained with her mouth open and a helmet with chin strap to stretch the connective tissues was applied at intervals during the day. Initial needle electromyographic study revealed that she had marked overall reduction of recruitment of motor unit potentials in the muscles of mastication with the left side significantly worse than the right. On repeat examination one week following her marcamine injection, the left masseter revealed a motor unit potential not seen on the previous examination. This finding may have been indicative of decreased activity of the antagonists that may have helped to recruit the agonist. Due to the improvement from the marcamine trial, she underwent botulinum toxin type A with ten units to each mylohyoid muscle under electromyographic guidance. She continued her oromandibular rehabilitation. At her two month outpatient follow-up she was able to actively close her jaw and was able remain closed at rest. On her last follow-up appointment she had made significant progress in physical, occupational, and speech and language therapies, where she was tolerating a regular diet and had started attending community college.

CONCLUSION

Botulinum type A toxin in conjunction with oromandibular rehabilitation, involving active soft tissue stretching passive positional assistance using a helmet with chinstrap to support the jaw, was used to treat jaw-opening oromandibular dystonia following her traumatic brain injury and mandibular fracture. This treatment allowed our patient to regain her speech and swallowing function along with improved cognition. The rehabilitation outcome became favorable after improved nutrition, speech with improved overall function.

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