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CASE REPORT

A case of asymmetric cranial nerve palsy due to iatrogenic botulism

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Abstract

Introduction: Botulism is a rare but lethal disease and atypical clinical presentations of this disease are difficult to diagnose. We report a case of iatrogenic botulism who presented withasymmetric cranial nerve involvement.
Case report: An eighteen year old female with cerebral palsy, congenital hydrocephalus and left upper/lower limb spasticity accidentally received double the normal dose of Botulinum neurotoxin A (BoNTA) which was equal to 800 Units, administered intramuscularly in the aforementioned muscle groups, leading to left eyelid drooping without no additional cranial nerve palsy or sensory/motor limb deficit. Patient was hospitalized for 3 days of clinical observation during which there was no progression of symptoms to the contralateral side, and the patient was uneventfully discharged. On follow up, the patient continued to make a steady recovery.
Discussion: Asymmetric muscle weakness is unusual for botulism confounded further in a patient with underlying spastic disorder. In our case, BoNTA was administered unilaterally in muscle groups, yet symptoms developed remotely from the injection site. Factors causing this could be large antero and retrograde axonal transport or from systemic uptake from nearby capillary beds. There is no current consensus on an optimal therapeutic injection dose for BoNTA in children or adults.
Conclusion: Physicians should be aware that asymmetric iatrogenic botulism of the cranial nerves may occur from a botulinum toxin injection into a site that is anatomically remote from the face.

Keywords: Botulism neurotoxin, Toxicology, Emergency, Antitoxin.

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INTRODUCTION

Botulinum neurotoxin A (BoNTA) has many applications within the fields of cosmetic surgery as well as neurology, dermatology, and gastroenterology. BoNTA prevents the release of acetylcholine at the endings of motor nerves leading to muscular relaxation. It has been acknowledged that this kind of treatment has a favorable safety profile and is generally well-tolerated in appropriate doses. Rarely, severe complications may occur with repeated or excessive dosages, which can include generalized muscle weakness, dysphagia, respiratory arrest, or death. Such disorders are referred to as iatrogenic botulism (include reference) for which a prompt clinical diagnosis is mandatory for early and effective treatment (1). According to a Botulism mortality survey in the US from 1975 – 2009, only five were reported as iatrogenic botulism (age range 34 to 89 yrs.) that received a highly concentrated, unlicensed preparation of BoNTA; but none of them died (2).

Asymmetric botulism is rarely reported due to a difficulty in clinical identification and infrequency of cases. Per a review article that examined clinical features in 55 food borne botulism patients, only few showed atypical presentations such as paresthesia (14%), asymmetric extremity weakness (17%) and asymmetric ptosis (8%) (3).

We report a case of iatrogenic botulism who presented with asymmetric cranial nerve involvement.

CASE PRESENTATION

An eighteen year old female with a previous medical history of cerebral palsy, congenital hydrocephalus requiring a ventriculoperitoneal (VP) shunt, and chronic left upper and left lower limb muscle spasticity requiring Botox therapy presented to the Emergency department (ED) with left eye ptosis.

According to the patient`s mother, she would regularly visit her neurologist who would inject a total of 400 Units of BoNTA administered intramuscularly into her the left distal biceps, pronater teres, tibialis posterior, medial/lateral gastrocnemius and soleus after which the patient would get slight relief in her spasticity and would engage in her normal daily activities. She developed left eye ptosis with no additional focal weakness or difficulty in breathing that was noted approximately one week after her most recent BoNTA

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injection which was an accidental double dose equal to 800 Units.

On arrival, she had a heart rate of 90 beats/min, blood pressure of 117/55 mmHg, respiratory rate of 18/min, oxygen saturation of 99% on room air and temperature of 37 degree celsius. Initial physical examination was significant for asymmetric left sided ptosis, without additional cranial nerve palsies. She had spastic tone in her left arm and leg with intact reflexes and strength. There were no sensory deficits.

Complete blood count, serum chemistry and cranial computed tomography were within normal limits, and the patient was hospitalized for 3 days out of concern for symptom progression. She was only monitored and remained stable without signs of respiratory distress or worsening cranial nerve palsies hence, was discharged. Four weeks after hospital discharge the ptosis had resolved.

DISCUSSION

Asymmetric muscle weakness is unusual for botulism but has been previously reported. Hughes et al. reported asymmetric findings in their review of 55 patients with foodborne botulism such as paresthesia (14%), asymmetric extremity weakness (17%) and asymmetric ptosis (8%) (3). Classically opthalmoplegia is a characteristic finding in however, Gdynia et al. reported a rare presentation of foodborne botulism with multiple bilateral cranial nerve palsies with sparing of estraocular musculature (4). Filozov et al. report a type F botulism case with asymmetric cranial nerve and extremity weakness with trunal ataxia (5). Such findings can be further confounded if a patient has an underlying spastic disorder as was seen in our patient.

In our case, BoNTA was administered unilaterally in muscle groups, yet the symptoms developed at a site anatomically remote from the injection site. The mechanism of distant transport is not clearly understood. Standard practice of aspirating for blood prior to injection likely excludes an inadvertent BoNTA injection into the systemic circulation. The remote effects can also result from large neuronal uptake of BoNTA with both anterograde and retrograde axonal transport as seen in rodent models or from its systemic distribution via nearby capillary bed uptake at the injection site (6-8). Factors that are usually considered to influence the risk of diffusion outside the target tissue include the characteristics of the preparation (e.g., the molecular size of the toxin), the dosing and injection technique, and the quality of muscle tissue itself (9,10).

There is no current consensus on an optimal injection dose for BoNTA in children or adults suffering from dystonias or limb spasticity. The exact dosage and the number of injections must be tailored to each individual, based on the size of muscle groups, number and location of the involved muscles, the severity of spasticity, the presence or absence of preexisting muscle weakness, and the patient's response to previous treatments (11). Finally, increased sensitivity to the toxin has also been theorized to effect dose-response to BoNTA. Our patient had received multiple prior injections without similar adverse reaction.

Botulism management, regardless of the exposure route, relies on supportive care in an intensive care setting with provision of mechanical ventilation, while awaiting the recovery of neuromuscular signaling mechanisms (11). Despite the absence of randomized trials, all data indicated that early antitoxin administration in severe or rapidly progressively cases could be lifesaving (12). Our patient did not exhibit progression of her symptoms and did not require treatment with antitoxin.

CONCLUSION

Providers should be aware that unilateral weakness can occur in muscle groups that are anatomically remote from a BoNTA injection site.

Conflict of Interest: None to be declared.
Finding and Support: None.

Ethical Statement: This research was conducted in accordance with the Declaration of Helsinki. Informed consent was obtained from the patient.

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