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Case Report

# Total Relief of Cluster Headache after Botulinum Toxin Injection: Case Report

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**Abstract**

A case of total relief of chronic cluster headache (CH) in a young male adult is reported on. 60 then 150U Botox* were used in an 18 year old male patient, presenting with chronic cluster headache, recalcitrant to conventional preventive treatment (Verapamil). Total relief, persisting after the second injection, was confirmed, and tolerability was excellent. Even if the pathophysiology of CH is not completely understood, use of botulinum toxin in such a condition would seem to be of therapeutic interest.

## INTRODUCTION

Cluster headache (CH) belongs to a group of primary headache disorders, classified as trigeminal autonomic cephalalgias. It is a neurological disorder characterized by recurrent, severe groups of headache attacks occurring together on one side of the head, typically around the eye. They are often accompanied by autonomic symptoms such as eye watering, nasal congestion and swelling of and around the eye, all confined to the side of the head with the pain [1].

The cause has not been identified, but the potential role of a dysfunction of the hypothalamus (which may explain why cluster headaches frequently strike around the same time each day, and during a particular season) seems to be well accepted after the works of PJ Goadsby [2]. While there is no cure, cluster headaches can sometimes be prevented and acute attacks treated. The recommended treatment for acute attacks is oxygen in combination with a fast acting triptan [3]. Primary prevention is founded on verapamil use. Steroids may be used to prevent a recurrence until verapamil takes effect.

Greater occipital nerve block has more recently been shown to be an effective alternative bridge therapy to oral medications, and thus Botulinum toxin type A (BoNT-A) has also been studied as a new preventive treatment for patients with chronic CH, with limited success [4,5]. Nevertheless, few studies are available and the real effect of toxin is still not totally known [6].

We report here on a single case of total relief of CH after botulinum toxin injection.

### Observation

An 18 year-old male was referred to us for chronic CH, running since 2010, partially relieved by Verapamil but with a tendency to be resistant to treatment. He had no remarkable past history, except for atopic background. He received on 08.20.2013, 60U of botulinum toxin A (Botox® / Allergan Pharmaceuticals, Westport, Ireland) in double dilution in the ipsilateralpericranium muscles and in topping the whole surface of cutaneous projection of the pain (Figure1).

At the first follow-up appointment, on 10.03.2013, the patient reported total relief of pain and disappearance of eye watering, with no need for verapamil. At the third appointment, on 11.27.2013, he complained of return of the attacks about 10 weeks after first injection. At the same time, he received a new injection of 150U Botox according to a standardised injection scheme, already used by us in other pathologies. This time relief was total and persisted at the last follow-up, allowing long relief expectations.

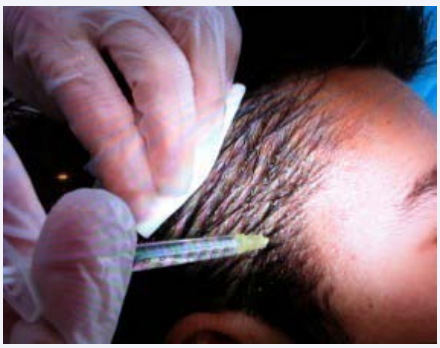

**Figure 1** Direct injection of BoNT-A by topping the whole surface of cutaneous projection of the pain.

## DISCUSSION

Botulinum toxin is the most powerful known neurotoxin, produced by an anaerobic gram-positive bacteria, *Clostridium botulinium*. This was described for the first time in 1817 by JC Kerner, but it was not until 1895 that the cause of botulism was highlighted by Van Ermengem, and the type A toxin was finally isolated by Sommer in 1920. Only after the Second World War did work on the different forms of toxin really begin, and their practical application into human pathology only began at the end of the 1970's with the work of Scott in the treatment of strabismus.The toxin acts by causing a sort of chemical denervation by blocking neurotransmitter release at the synaptic cleft of the acetylcholine fibers of the motor nerves and of the autonomic nervous system [7], and studies at the start of the 2000's have demonstrated that other neurotransmitters are blocked by BoNT-A too [8]. Contrary to other drugs such as atropine or probanthine, there are few side effects, and these are restricted most of the time to the area of injection (allergic reaction or muscular weakness) [9].

Botulinum toxin has already been successfully used in the treatment of spasmodic torticollis [10], in laryngeal dystonia, hemifacial spasm, blepharospasm, andin different types of painful pathologies for a few years [11,12]. Our team has studied two other pathologies treated with BoNT-A: trigeminal neuralgia (13) and occipital neuralgia [14].

An open label study on the efficacy of the toxin in refractory chronic tension-type headache, showing significant reductions in headache frequency, intensity and analgesic consumption which persisted up to 1 year, came in 2007 [15].

Interest of its use in CH seems to go back to the same period, when Sostak and et al. [6], report on an open study using botulinum toxin in 12 patients, either with episodic or chronic presentation, all unresponsive to common prophylactic medications. In one case of chronic presentation, total relief was obtained, and significant relief was obtained in two other chronic cases of CH in young male patients. Interestingly, no significant relief was shown in any case of episodic presentation.To note is the use of only 50U Botox® by injection in the standardized scheme reported in their study. In our case, the presentation was strictly the same, concerning a young male patient with chronic presentation, but with prolonged relief obtained only after increase of the doses, i.e.150U at the second injection.

To the best of our knowledge, no other large studies have been published on the question, even if occasionally its use is mentioned in a trivial way [5,16].

In our case, as in other pathologies, the use of botulinum toxin was safe and does not produce systemic effects associated with other types of headache treatment.

## CONCLUSIONS

Although the pathophysiology of CH is incompletely understood, muscle tension may trigger or aggravate headaches. Botulinum toxin, which reduces muscle hyperactivity, may reduce headache pain by decreasing muscle tension. It also provides peripheral and central neurogenic effects and reduces inflammation. Large, rigorously controlled trials of botulinum toxinare needed to better characterize its role as a potential therapeutic agent.

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