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An unusual complication of Botox treatment for sialorrhoea



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ABSTRACT

INTRODUCTION: To illustrate the potential side effects and clinical efficacy of Botox injections in treating sialorrhoea.

PRESENTATION OF CASE: A 26-year-old patient with cerebral palsy with dystonia had a long history of severe, distressing sialorrhoea. She was treated with three separate Botox injections into her salivary glands in December 2011, July 2012 and March 2013.

DISCUSSION: Following the Botox injections the patient developed dysphagia, began to expectorate thick mucus and developed a cough; she was treated for a chest infection and during this time her feeding deteriorated. Three injections were given as the patient had an objective and significant reduction in salivation. However, the side effect profile was deemed too great to continue with treatment.

CONCLUSION: Botox is a novel and effective treatment for reducing saliva production. Its clinical efficacy is supported by this case and correlates with the recent literature. Although rare, significant side effects can happen and the case presented illustrates the care needed when administering injections, particularly in a subgroup of patients.

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1. Introduction

Pathological sialorrhoea can be due to intrinsic hypersalivation or occur secondary to neurological disorders such as amyotrophic lateral sclerosis (ALS), cerebral palsy (CP), Parkinson disease (PD), or as a side effect of medications.¹ It can cause both psychological and physiological distress to patients. In children, the most common cause of sialorrhoea is CP with hypersalivation occurring in 10–38% of patients.² In adults, PD is the most common cause with 70–80% of PD patients demonstrating sialorrhoea.³ Sialorrhoea resulting from neurological conditions has a complex aetiology and is produced by impaired swallowing as a result of compromised neuromuscular function and aberrant coordination of the oral, facial and lingual musculature.¹

Botulinum toxin (BoNT) is a neurotoxin that blocks the release of acetylcholine and a number of other neurotransmitters from synaptic vesicles.⁴ Its effect on drooling was first noticed in patients with Parkinson disease and has been reported to be effective in the treatment of sialorrhoea in numerous case studies, clinical trials and retrospective studies.⁴ At present, three type A and one type B toxins are in clinical use; onabotulinumtoxinA, abobotulinumtoxinA, incobotulinumtoxinA, and rimabotulinumtoxinB.¹

2. Case report

A 26-year-old patient with cerebral palsy had suffered from sialorrhoea all her life. She was treated with three separate Botox injections into her salivary glands in December 2011, July 2012 and March 2013. After the first Botox injection she developed breathing difficulties, a nocturnal cough, produced thick mucus and seemed to choke whilst eating, suffering from dysphagia. Two courses of antibiotics were given, but they had no clinical effect. The patient gradually improved, but her appetite was still reduced.

After the second injection the patient's feeding deteriorated again and she developed dysphagia and a distaste of her food. Also, she started to produce more thick mucus and choked at night. She was assessed by a speech and language therapist who advised a soft mash/pureed diet with thickened drinks. In addition to this she was prescribed carbocysteine that did have some effect. Over the next few months her feeding proved very difficult with frequent choking episodes and a general distaste for food leading to a reduced appetite and weight loss.

Despite the side effects listed above the Botox injection were effective at treating the patient's sialorrhoea and she found this socially more acceptable and comfortable. Therefore a further Botox injection was administered. Within three days her feeding deteriorated alarmingly as noted by her parents. She began choking on all food and drinks, with severe episodes making her nauseous and pale. Moreover, taking her oral medication was difficult and she was expectorating thick viscous mucus.

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The parents of the patient declared that there was no doubt in their minds that the side effects described above were associated with the administration of Botox into her salivary glands.

3. Discussion

BoNT is an established treatment option to treat hypersalivation. Fuster Torres et al.⁵ stated that a significant reduction was observed in the production of saliva following injections in a number of studies and the duration of the therapeutic effect was 1.5–6 months.⁵ Hay and Penn⁶ also noted a statistically significant decrease in drooling in a paediatric population.⁵ The case presented here validates the clinical efficacy of Botox in treating sialorrhoea and the patient's carers acknowledged an objective reduction in drooling.

Botox injections are a less invasive treatment strategy for sialorrhoea compared to surgery. Various surgical procedures are available, such as salivary gland duct ligation that is usually used as a last resort when other methods have failed and in severe refractory cases. Behaviour modification techniques, correction of situational factors and oral-motor therapy are other treatment options, but do not always produce a consistent improvement. Oral agents, such as glycopyrrolate, and topical agents are also offered with varying degrees of success. A potential disadvantage of Botox injections is the array of side effects that can be encountered as illustrated in this case. However, in a large scale study Amrita Lakraj et al.¹ defined side effects as uncommon and manageable when injections were delivered at experienced centres.

Chan et al.⁷ recently performed a review of the long-term safety and efficacy of Botox injections for sialorrhoea in 69 children. They declared that postinjection complications occurred in 19 patients (23 events). This was fourteen patients (15 events) with minor and five (8 events) with major complications. Major complications included aspiration pneumonia ($n = 3$), severe dysphagia ($n = 2$), and loss of motor control of the head ($n = 3$), resulting in 5 hospitalizations and 2 nasogastric tube insertions. The patient in our report did suffer from dysphagia, and possibly a chest infection, but did not require hospitalization.

In contrast, Amrita Lakraj et al.¹ concluded that along with a significant positive clinical effect in patients BoNT had a limited side effect profile. They stated hypersalivation in 3.9%, dysphagia in 3.3%, xerostomia in 3.3% and pneumonia in 2.2% of patients. The side effects listed here are only apparent in a small minority of the patient sample, but the adverse effect seem characteristic.

As Botox injections are frequently used in patients with pre-existing dysphagia and other oropharyngeal neurological deficits the side effect of dysphagia may be significant. Any exacerbation of swallowing difficulties could have the potential to cause aspiration pneumonia.⁸ Dysphagia may result from inaccurate injection of the Botox into the salivary glands signifying the importance of localizing the correct site before the drug is administered.

4. Conclusions

Numerous studies validate the clinical efficacy of Botox injections in sialorrhoea and the patient presented here supports this data. There is also a wealth of information describing potential side effects of Botox injections, but these are only experienced in a minority of patients. This case is an unusual example of significant side effects resulting from Botox treatment. Also, the cardinal side effects experienced by the patient correlate with reported side effects in the literature, notably dysphagia, increased mucus expectoration and possibly a chest infection. This illustrates the need to consider possible side effects when giving Botox injections as they have the potential to cause significant morbidity, particularly in patients with severe neurological disorders.

Conflict of interest

The authors declare that they have no conflict of interest.

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Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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