

# Botulinum Toxin in the Treatment of Lingual Dystonia Induced by Speaking

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## Key Words

Lingual dystonia · Botulinum toxin · Speaking

## Abstract

Primary lingual dystonia is a rare condition, especially when it is only induced by speaking. Trihexyphenidyl failed to improve the symptoms. Several case series have demonstrated the effectiveness of botulinum toxin injection for the management of focal lingual movement disorders. Only 1 case of botulinum toxin injection for primary lingual dystonia induced by speaking has been reported, but this treatment has limited effectiveness. Our patient was treated with botulinum toxin using a superficial approach for injection into the tongue with continuing excellent results. Lingual botulinum toxin injection is a fairly simple, safe and viable treatment option for lingual dystonia induced by speaking.

## Introduction

Primary lingual dystonia is a rare condition, especially when it is only induced by speaking [1–6]. There are a few papers in the literature reporting on experience with botulinum toxin for lingual dystonia [7–11]. We describe a 69-year-old female patient with lingual dystonia treated with botulinum toxin using a superficial approach for injection into the tongue.

## Case Report

The patient is a 69-year-old woman, previously well with no significant medical or family history, who presented with gradual difficulty in speaking over a period of a few

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months. Her tongue would protrude and occasionally pulled to 1 side of the mouth while speaking. She noticed that her tongue spasms and contortions only occurred during speaking but not during other tasks such as eating, drinking, whistling, and blowing. Her speech rate was markedly reduced and she was hardly intelligible (online suppl. video 1, for all online suppl. material, see [www.karger.com/doi/10.1159/000347000](http://www.karger.com/doi/10.1159/000347000)).

She found that chewing bubble gum effectively relieved her symptoms. However, this sensory trick only lasted for a few weeks, followed by a gradual return of her symptoms. There was no history of exposure to neuroleptic medications, facial or oromandibular injuries, or recent infection. There were no involuntary movements of the neck or extremities. Her gait, extraocular movements, visual fields, strength, senses, cerebellum and reflex testing were normal.

Results of cranial and cervical magnetic resonance imaging, electroencephalogram, full blood count, serum chemistry, thyroid tests, serum ceruloplasmin level and 24-hour urinary copper excretion were all normal. Acanthocytes were not observed in the peripheral blood smear. She was treated with trihexyphenidyl (5 mg/day) with little effect.

Subsequently, the patient was treated with botulinum toxin injections in 2 points of each genioglossus, starting with 5 U and escalating to 10 U. Botulinum toxin was injected into the genioglossus muscle through an approach directly via the superior aspect of the tongue. She was injected every 3 months for 2 years with continuing excellent results. The only adverse effect was a mild swallowing difficulty that lasted for 2 weeks. The patient has remained asymptomatic 1 year later (online suppl. video 2).

## Discussion

Primary lingual dystonia induced by speaking is a rare disorder in which increased tonus of the tongue causes forced protrusion only during speaking. The literature includes 6 cases (7 including our patient) of primary lingual dystonia induced by speaking [1–6].

One important feature of cases with lingual dystonia is responsiveness to sensory tricks [1, 2, 4, 5], for example, how chewing gum effectively relieved symptoms in our patient. However, this sensory trick only lasted for a few weeks, after which she experienced a gradual return of her symptoms.

Our patient had no history of exposure to neuroleptic medications, facial or oromandibular injuries, or recent infection. For this reason, the condition was diagnosed as a lingual dystonia specifically induced by speaking after all laboratory examinations had ruled out secondary causes. The pathophysiologic mechanism of primary lingual dystonia induced by speaking remains unclear, and often the treatment of these patients is difficult. Some authors have suggested that anticholinergic medication, such as trihexyphenidyl, may be the treatment of choice in cases with lingual dystonia [1–3, 6]. In contrast to these cases, we observed that primary lingual dystonia may be intractable to anticholinergic treatment.

Several case series have demonstrated the effectiveness of botulinum toxin injection for the management of focal lingual movement disorders [7–11]. Only 1 case [5] of botulinum toxin injection for primary lingual dystonia induced by speaking has been reported, but this treatment has limited effectiveness.

The genioglossus makes up the bulk of the tongue's muscle mass and is responsible for tongue protrusion, making it an ideal candidate for injection. Recently, a report described an effective alternative approach with direct injection of botulinum toxin through the superior aspect of the tongue [10]. We chose the intraoral approach because of its simplicity and obvious accuracy since the genioglossus muscle is easily accessible. In our patient as well as

in several other cases with lingual dystonia, the average dose (20–30 U BTX-A) of botulinum toxin was similar, and there was a 15-week average duration of benefit [7–11]. The only adverse effect was a mild swallowing difficulty. Lingual botulinum toxin injection is a fairly simple, safe and viable treatment option for lingual dystonia induced by speaking.

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