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Review article

Relevance of intraglandular injections of Botulinum toxin for the treatment of sialorrhea in children with cerebral palsy: A review

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ABSTRACT

Background: After the age of 4 years, drooling becomes pathological and impacts the quality of life of children with cerebral palsy. Intraglandular injection of Botulinum toxin is one of the treatments available to limit this phenomenon.
Aims: The objectives of this review were to validate the efficacy of Botulinum toxin injections for drooling in children with cerebral palsy, determine recommendations and identify potential side effects.
Methods: We conducted a literature review from 2001 in the following databases: Embase, Pubmed and Cochrane using the keywords: sialorrhea, drooling, hypersalivation, Botulinum toxin, cerebral palsy and children. Only the articles evaluating the efficacy of Botulinum toxin in children with cerebral palsy over the age of 4 were researched.
Results: Eight studies were found: 2 case studies, 3 open and non-controlled studies and 3 randomized controlled trials. Efficacy results in this indication are quite encouraging and the use of BTX injections is safe but the overall level of evidence of these studies was quite low.
Conclusion: However, intraglandular injection of Botulinum toxin has a place among the therapeutic array available for the management of sialorrhea in this population even if no standardized protocol is available yet.

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1. Introduction

Children prior to puberty produce between 750 and 900 mL of saliva per day, reaching 1000–1500 mL in adulthood.[1] Apart from mealtimes, about 75% of saliva is produced by the submandibular glands as a mixture of both serous fluid and mucus, vs. 20% produced by the parotid glands as serous fluid. When chewing food, this ratio becomes reversed.[2,3] The regulation of this production is managed by the autonomic nervous system; its quantity is controlled by the parasympathetic system and its quality by the sympathetic system.

Sialorrhea, also called ptyalism or drooling, is defined as unintentional loss of saliva; when severe it is referred as anterior drooling. It can lead to facial skin irritations, halitosis, infections of the oral mucosa and perioral region, tooth decays, hygiene issues or dehydration. Furthermore, excessive drooling can damage the computers, technical aids or communication systems used by these patients. Socially, it has a negative impact since children need to wear a bib and have their clothes and/or bibs changed regularly.

Anterior drooling should be distinguished from posterior drooling, in which saliva can be inhaled and cause repeated pneumonia.[1] Regardless of its type, drooling can put an additional burden to the disability of these patients, with lowered self-esteem and social isolation.[4–6]

Drooling concerns 10–57% of children with cerebral palsy, the highest rates were found for children in institutions and for 33% of them the drooling was deemed severe.[7]

The reported underlying mechanisms of sialorrhea are not unequivocal: it can be due to increased saliva production, or hypersalivation, because of hypertrophied salivary glands, as observed during surgery, as well as inefficient oral neuromuscular control preventing complete deglutition. This impairment could be related to a defect in lip closure and lip control, tongue thrusting and jaw positioning, poor coordination of the orbicularis oris and masseter or dental malocclusion. In children with cerebral palsy, the deglutition reflex can sometimes be intact but swallowing frequency is reduced thus leading to saliva stagnation. Some authors have also brought up the potential role of reduced intra-oral sensitivity.[6,8] The disorders observed could also be related to an affection of the primary motor and sensitive areas as well as the anterior cingulate cortex.[9]

Some factors have been shown to promote drooling: poor head control or gastroesophageal reflux disease (GERD). Furthermore, spastic quadriplegia seems a greater purveyor of sialorrhea than diplegia or athetosis. However, not all authors have validated a correlation between drooling and altered cognitive functions.[7,10,11]

Some extrinsic promoting factors have been reported: antiseizure medications, acetylcholinesterase inhibitors, but also benign ENT infections of the upper respiratory tract, tooth decays, finger chewing or the patient's emotional state.[6,11]

Apart from controlling potential promoting factors, several therapeutic methods can be proposed to manage drooling in children with cerebral palsy:

- non-invasive methods: oral-motor therapy and medications (muscarinic receptor antagonists),
- invasive methods: surgery,
- other semi-invasive methods: acupuncture, photocoagulation or Botulinum toxin injections.

In 2001, Jongerius et al. published a first case study on children with cerebral palsy suffering from severe drooling treated by Botulinum toxin (BTX) injections.[12] Its antisecretory mechanism blocks the parasympathetic command of saliva production. Some reviews have evaluated the efficacy of BTX but in heterogeneous groups, made up of children and adults with different neurological and behavioral disorders.[13,14] Since 2001, several more studies on intraglandular BTX injections in children have enriched the literature but again covering wide etiological frameworks.[4,15–18]