

Abstract Botulinum toxin injections after surgery in Hirschsprungs disease: a systematic review and meta-analysis

Background

A large proportion of patients with Hirschsprung disease experience persistent obstructive symptoms after corrective surgery. Persistent obstructive symptoms may result in faecal stasis that can develop into Hirschsprung-associated enterocolitis, a potential life-threatening condition. Important treatment to improve faecal passage is internal anal sphincter relaxation using Botulinum toxin injections.

Aim

To give an overview of all empirical evidence on the effectiveness of Botulinum toxin injections in patients with Hirschsprung disease.

Methods

A systematic review and meta-analysis was done by searching Pubmed, Embase and the Cochrane Library, using entry terms related to (1) Hirschsprung disease and (2) Botulinum toxin injections. 14 studies representing 278 patients met eligibility criteria. Data that were extracted were proportion of patients with improvement of obstructive symptoms or less enterocolitis after injection, proportion of patients with adverse effects and data on type Botulinum toxin, mean dose, average age at first injection and patients with associated syndromes. Random-effects meta-analysis was used to aggregate effects and random-effects meta-regression was used to test for possible confounding factors.

Results

Botulinum toxin injections are effective in treating obstructive symptoms in on average 66% of patients (ER=0.66, $p=0.004$, $I^2=49.5$, $n=278$ patients). Type of Botulinum toxin, average dose, average age at first injections and proportion of patients with associated syndromes were not predictive for this effect. Mean 7duration of improvement after one BTI was 6.4 months and patients needed on average 2.6 procedures. There was a significant higher response rate within one month after Botulinum toxin injections compared to more than one month after Botulinum toxin injections (ER=0.79, *vs.* ER=0.46, $Q=19.37$, $p<0.001$). Botulinum toxin injections were not effective in treating

enterocolitis (ER 0.58, $p=0.65$, $I^2=71.0$, $n=52$ patients). There were adverse effects in on average 17% of patients (ER=0.17, $p<0.001$, $I^2=52.1$, $n=187$ patients), varying from temporary incontinence to mild anal pain.

Conclusion

Findings from this systematic review and meta-analysis indicate that Botulinum toxin injections are effective in treating obstructive symptoms and that adverse effects were present, but mild and temporary.