CASE REPORT

Two Cases of Severe Non-specific Oesophageal Dysmotility Showing Different Response to Botulinum Injection Therapy

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Summary

We report 2 cases where treatment of achalasia type symptoms due to severe non-specific oesophageal dysmotility have shown symptom resolution and manometric improvement to intrasphincteric botulinum injections either by itself or in combination with oesophageal dilatation.

Key Words: Botulinum toxin, Non specific oesophageal dysmotility

Case Report 1

A 47 year old lady presented with recurrent vomiting. There was a past history of haemetemesis in 1987 due to a prepyloric gastric ulcer and another episode in 1988 secondary to a Mallory-Weiss tear. A hiatus hernia was diagnosed in 1989 on endoscopy. In 1995 she presented with vomitting and abdominal pain. An upper gastrointestinal endoscopy was normal and she was discharged the next day. She continued to have recurrent vomiting and sought treatment from her general practitioner with little symptomatic improvement. She was readmitted in April 1998 with haemetemesis and melaena. Upper GI endoscopy showed a Mallory Weiss tear and fresh blood in a large hiatus hernia. During this admission, she continued to have persistent vomiting. A thoracic and abdominal CT scan failed to reveal mechanical obstruction of the gastrointestinal system. An abdominal ultrasound and barium meal and follow through were also normal. Oesophageal manometry revealed a lower oesophageal pressure of 25mmHg (normal range 6 - 15mmHg) with no evidence of relaxation to swallowing. 20% of contractions in the distal oesophagus were non-propulsive, the findings were consistent with a severe non specific motility disorder. As some of these patients progress to a more classical disorder such as early or vigorous achalasia an upper GI endoscopy was performed and botulinum injection was administered (20 units each) in each of four quadrants at the lower oesophageal junction with a sclerotherapy needle. The patient was continued on proton pump inhibitor therapy after the procedure. On review a month later she reported feeling much better and did not have recurrent episodes of vomiting. Repeat manometry almost 5 months after the botulinum injections indicated marked improvement. The lower oesophageal pressure had reduced to 11mmHg and there was complete relaxation of the sphincter to swallowing and no residual pressure noted. The oesophageal body motility showed monophasic contractions, co-ordinated motility and resolution of the previously noted non-specific motility disorder in the oesophageal body. The patient was totally asymptomatic. This was possibly also

helped by the fact that she had managed to lose weight. This may additionally reflect an improved sense of well being and motivation.

Case Report 2

The second patient was a 28 year old lady who presented with a history of several months of dysphagia in August 1997. She complained that food, in particular solids, tended to stick in her chest on swallowing, and was sometimes associated with retrosternal and epigastric pain. She had lost weight since the onset of symptoms. Her initial upper GI endoscopy showed initial resistance to the passage of the scope at the lower end of the oesophagus. *Helicobacter pylori* testing was positive for which she was given a course of triple eradication therapy. Oesophageal manometry assessment showed lower oesophageal sphincter pressures of 17mmHg (Normal range 6 - 15mmHg) but only partial relaxation of the lower oesophageal sphincter to swallowing at 9mmHg. There were simultaneous contractions and an increased mean duration of contractions to 8 seconds which were consistent with ineffective oesophageal motility. A significant non-specific motility disorder was diagnosed and in view of the possibility of progression to a more classical achalasia type disease, 20 units of botulinum toxin injections was injected each into 4 quadrants at the lower oesophageal sphincter with a sclerotherapy needle at upper gastrointestinal endoscopy. When reviewed 3 weeks later in the clinic she reported remarkable clinical improvement with good ability to swallow accompanied by some weight gain. Repeat oesophageal manometry showed a decrease in the lower oesophageal pressure (13mmHg) with improved relaxation to 5mmHg. Although there was still a residual body motility disorder, there was an improvement in peak amplitudes of contraction. At 3 months she had a recurrence of symptoms and underwent another course of botulinum toxin injections, resulting in symptomatic relief and weight gain. There was significant improvement of her lower oesophageal sphincter pressures on oesophageal manometry. Despite this initial remarkable improvement in symptoms, she had recurrence within 3 months and subsequently underwent oesophageal dilatation, which failed to alleviate her symptoms. This was followed by another dilatation combined with botulinum injection, 3 months later. Since then she has had significant symptom resolution and her manometry confirmed improved sphincter pressure. This has persisted for 4 months.

Discussion

The first case highlights the therapeutic efficacy of botulinum toxin injections for this disorder. Both patients have shown sustained clinical improvement and this has been objectively documented at manometry. Their lower oesophageal pressures have shown a fall of over 50% and the achalasia type non-specific motility of the oesophageal body resolved dramatically. As Case 1 is not a very suitable candidate for surgery in view of obesity, this has proven to be an effective alternative therapy. It is also cost effective.

This is the first case report of a patient with achalasia type non-specific motility and a large hiatus hernia. We are not certain if there exists a definite link between these two conditions. There may however be a causal link between the repeated episodes of lower oesophageal mucosal injury (Mallory Weiss tear) and her symptoms.

The second patient proved to be less responsive to either botulinum injection or oesophageal dilatation on its own. However, a combination of the two has resulted in a rather more prolonged remission but this may need to be repeated at a future date if recurrence occurs. We acknowledge that repeated injections might be needed in order to achieve sustained remission. It has been shown that a repeat course of botulinum injection can aid in prolonging further relapse of symptoms. These patients will require long term follow up and it is an acceptable option in non-surgical patients. Its use has also been suggested in some malnourished patients to attain an adequate nutritional status before surgery for achalasia.

The side effect profile of botulinum toxin injection therapy is generally encouraging and it is a safe and simple method of treatment of achalasia[1]. In a series by Pasricha *et al*[2], no serious adverse effects were encountered in 21 patients who underwent this treatment. In conclusion, botulinum toxin either on its own or in combination with oesophageal dilatation is useful in treating 'achalasia-like' oesophageal dysmotility.

References

1. Schiano TD, Fisher RS, Parkman HP, Cohen S, Dabezies M, Miller LS: Use of high-resolution endoscopic ultrasonography to assess oesophageal wall damage after pneumatic dilatation and botulinum toxin to treat achalasia, Gastrointest Endosc, 1996; 44: 151-7.

2. Pasricha PJ, Rai R, Ravich WJ, Hendrix TR, Kalloo AN: Botulinum toxin for achalasia: Long-term outcome and predictors of response: Gastroenterology, 1996; 110: 1410-5.