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Clinical Record

Botulinum neurotoxin for management of intractable central leakage through a voice prosthesis in surgical voice restoration

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Abstract

Rehabilitation of voice and speech after total laryngectomy has become established practice in recent years. A voice prosthesis is placed within a surgically produced fistula between the trachea and upper oesophagus and acts as a one way valve, allowing passage of pulmonary air from the trachea into the oesophagus and preventing aspiration of food and fluid from the oesophagus into the trachea. Persistent leakage through or around these prostheses is a recognized complication, the aetiology of which can vary widely, from mechanical issues with the prostheses themselves to anatomical and physiological issues associated with the reconstructed pharynx.

We report a new technique of using Dysport® in the management of intractable central leakage due to premature and forceful closure of the upper oesophageal sphincter during swallowing. This resulted in the pooling of fluids around the posterior flange of the prosthesis. This, along with the increased pressure from the muscle contraction, led to central leakage, as identified on videofluoroscopy. An injection of Dysport paralysed the upper oesophageal sphincter, preventing pooling of fluids around the prosthesis and the forcing open of the valve. The effect was to eliminate the leakage, and the patient did not require further injections over the following 22 months.

Key words: Botulinum Toxin Type A; Laryngectomy; Voice Quality; Fistula; Larynx, Artificial

Introduction

In 1979, Mark Singer and Eric Blom first established the technique of tracheoesophageal puncture (TEP) for surgical voice restoration.[1] Since that time, it has become an established procedure in the rehabilitation of voice and speech after total laryngectomy.

There are various types of prostheses available commercially, all of which operate on the common principle of a one way valve which allows passage of pulmonary air from the trachea into the oesophagus and prevents aspiration of food and fluid from the oesophagus into the trachea.[2] These prostheses are constructed of medical grade silicon and are placed within the TEP by either an ENT surgeon or a speech and language therapist. There are a number of inherent problems with use of a device such as this within the tracheoesophageal party wall. One of the most common management problems is leakage of liquid through the prosthesis in situ. This usually indicates failure of the valve mechanism and requires a new valve to be fitted. The common causes are related to age of the prosthesis or overgrowth of fungal infection on the prosthesis itself.[3,4] This case study addresses a central leakage in the absence of fungal growth or valve deterioration.

Injection of botulinum neurotoxin is a recognized technique[5] for pharyngeal constrictor muscle spasm in tracheoesophageal voice restoration and is a technique we have used with success.[6] This article is the first to report the use of Dysport in the management of intractable central leakage associated with swallowing, in a case in which voice production was unaffected.

Case report

A 55-year-old man underwent total laryngectomy with primary tracheoesophageal puncture and myotomy for carcinoma of the larynx in October 2001. He achieved tonic voice quality with a 14 mm low-pressure Blom–Singer voice prosthesis, which he maintained throughout his postoperative radiotherapy. A low-pressure prosthesis was selected due to its minimal profile within the oesophagus. In April 2002, the tracheoesophageal party wall became oedematous, pulling the prosthesis into the tract. As a result, the posterior end of the TEP closed and the patient required a secondary procedure.

Following the secondary procedure, the patient experienced intermittent central leakage, using the same type of low-pressure prosthesis but down-sized to 8 mm. A number of recognized techniques were tried, including: checking the patient's cleaning techniques in situ; reinforcing the importance of weekly alternating and changing and the use of disinfecting procedures between two prostheses; and commencement of a standard itraconazole regimen. However, the intermittent central leakage continued.

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Examination under videofluoroscopy initially failed to demonstrate the leakage reported by the patient; maximum dilation of the reconstructed pharynx was achieved, no spasm was evident and the flapper valve did not open on inspiration. We hypothesized that the flapper valve within the prosthesis may have been intermittently pulling open during swallowing, outside the clinic setting. The patient was subsequently switched to a hooded valve (8 mm), which had a recessed flapper valve less likely to be affected by pressure variables during oropharyngeal swallow. This initially solved the central leakage, and we concluded that our hypothesis was correct. However, after four months, the central leakage problem recurred in the absence of fungal growth on the prosthesis or any further obvious oedematous changes to the party wall. There had never been any peripheral leakage.

A second videofluoroscopy was performed to assess any physiological changes. Results demonstrated the bolus passing freely into the neo-pharynx, over the posterior flange of the prosthesis and into the oesophagus, with maximum dilation of the neo-pharynx. The upper oesophagus just inferior to the prosthesis closed prematurely, with back-flow of part of the bolus occurring after every third or fourth swallow, resulting in pooling inferior to the prosthesis. The back-flow proceeded in a south–north direction and hence could flow under the hood of the valve into the barrel of the prosthesis. Use of a duckbill prosthesis remained impossible due to its extended profile into the oesophageal lumen, resulting in splaying against the posterior oesophageal wall.

Figure 1 shows spasm of the upper oesophagus, causing bolus to flow in a cranial direction, with subsequent pooling around and under the hood of the prosthesis.

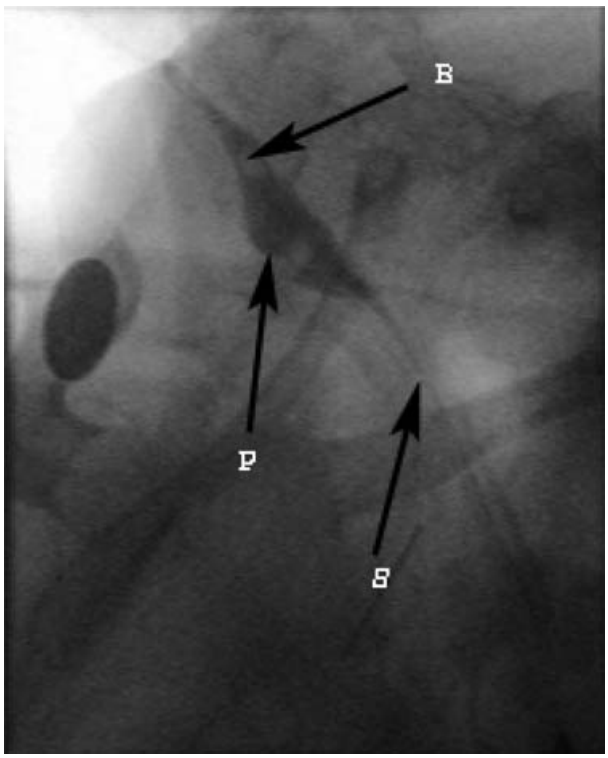

FIG. 1

Videofluoroscopy showing spasm of the upper oesophagus, causing bolus to flow in a cranial direction with subsequent pooling around and under the hood of the prosthesis. S = area of spasm; P = pooling; B = area of back-flow under the prosthesis, causing the central leakage

We felt that elimination of this premature contraction would allow the bolus to pass the valve without the consequent leakage, so we injected 250 units of neurotoxin (Dysport) (a similar amount to that used for spasm in voice restoration) into the posterior wall of the upper oesophagus just below the level of the puncture.

Due to our extensive experience with using Botox in laryngectomy patients for voice restoration purposes, we performed the procedure in the same manner as for patients undergoing injection for voice restoration,[7] that is, in the out-patients department, without the need for electromyography.

The procedure was achieved by injecting just superolateral to the stoma and angling a 23 gauge needle down and medially (see Figure 2). On feeling the prevertebral muscles, which have a characteristic 'crunchy' feel, the needle was withdrawn back into the posterior oesophageal wall and Dysport (diluted to 3 mls) injected over a 2–3 cm length of muscle.

The patient was sent home and asked to monitor central leakage, record his cleaning and changing regime, and report any changes in voice quality. Results were excellent, and subsequent videofluoroscopy six weeks after the procedure indicated that the premature closure of the upper oesophagus had significantly changed. There was no longer evidence of back-flow of part of the bolus (which had previously resulted in pooling beneath the prosthesis), as can be seen in Figure 3.

The position of the posterior flange had not changed with respect to the oesophageal wall and there was no change in voice quality. The procedure also resulted in a widening of the oesophageal lumen, which allowed placement of the duckbill prosthesis, with no splaying against the posterior oesophageal wall. This prosthesis was more durable, and less likely to be dislodged from the TEP by such actions as coughing, than other, comparable prostheses with a lower oesophageal profile.

Twenty-two months after the procedure, the patient was still wearing a Blom–Singer duckbill voice prosthesis. He was completely independent in the care and management of his voice prosthesis. Cleaning, changing and disinfecting routines had been reinstated, prostheses were lasting four to six months and the patient no longer required an antifungal regime. The leakage issues had been resolved, with no compromise of voice quality. The patient no

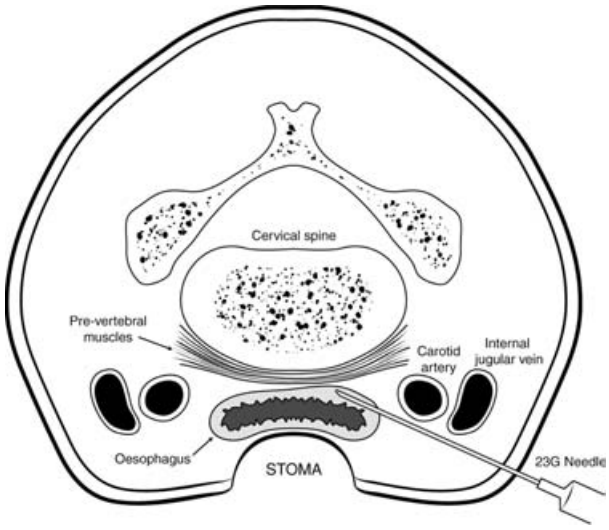

FIG. 2

The path of needle introduction for lignocaine/botox injection.

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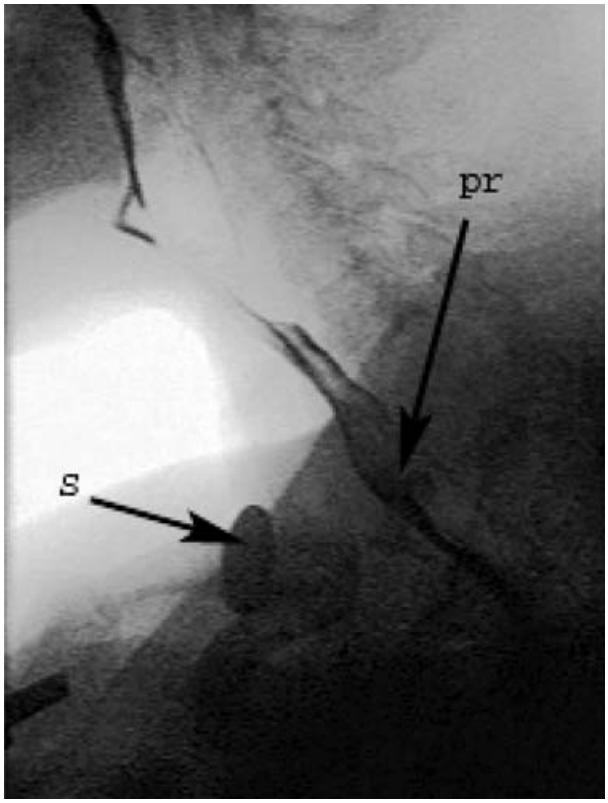

FIG. 3
Videofluoroscopy; S shows the stoma site (marked by a coin); Pr indicates the new prosthesis alignment (note that there is no longer any evidence of pooling or spasm).

longer needed special anti-fungal regimes (apart from the routine hygiene practices all patients with a valve need to undertake).

Discussion

Central leakage is most commonly caused by fungal growth on the flapper valve of the prosthesis itself.[3] This is not an unexpected problem but varies in its degree and ease of management. Central leakage also occurs for other reasons, related to pressure changes in the neo-pharynx during swallowing, as outlined in this paper. Regardless of the source of the central leakage, it must be treated as a potentially serious problem as it can lead to aspiration and chest infection.

It is well established that radiotherapy and a thin party wall can lead to leakage around a prosthesis;[8] however, this did not contribute to leakage in our patient.

Botulinum neurotoxin binds to the motor nerve terminal and therefore needs to be delivered into the muscle and, unlike lignocaine, will have no effect on the neural plexus outside the muscle of the nerve terminal. Botulinum neurotoxin serotype A blocks acetylcholine release by cleaving a cytoplasmic protein that is located on the cell membrane and required for the release of this transmitter. The affected terminals are inhibited from stimulating muscle contraction. The toxin does not affect the synthesis or storage of acetylcholine or the conduction of electrical signals along the nerve fibre.[9]

Over time, muscle activity gradually returns, either by growth stimulation of collateral axonal sprouts or, as new research suggests, by the return to functionality of the original junction.[9]

In most clinical situations, therefore, repeat injections of botulinum are usually required to maintain the desired clinical effect.

Post-laryngectomy botulinum neurotoxin is most commonly used within the reconstructed pharynx to achieve tonic voice production when this is compromised by spasm or hypertonicity of the mucosal vibrating segment.[7] Contrary to expectation, however, in 66 to 85 per cent of cases in which botox was injected to eliminate spasm during voicing, only one injection was necessary for patients to maintain their voice long term (personal series – P M Clarke, unpublished data). The majority of the remaining patients required only one additional injection to maintain long-term effects. One hypothesis for why the effect of botulinum lasts longer than expected is the slow process of recovery that allows the previously hypertonic segment to be 'retrained' to its new usage as a vibrating segment. Hence, over time, the muscle is less likely to spasm when stimulated by airflow during voice production. The elimination of spasm, evidenced by the ongoing production of voice as well as by videofluoroscopy results, is maintained long after the effect of botulinum would have been reversed.

- **Central valve leakage can be a difficult complication of speaking devices**
- **In a small number of cases, this is due to premature or hypertonic closure of the upper oesophageal sphincter**
- **Botulinum neurotoxin has been used in hypertonic spasm to restore voice, with good effect**
- **In central leakages due to spasm, injection with botulinum toxin may be a new, straightforward option**

Using botulinum neurotoxin in this way allowed us to manage an intractable and potentially serious problem for our patient. Once the problem had been diagnosed, the procedure was straightforward and was carried out in the out-patient department under local anaesthetic.

The problem did not recur over the 10 months following the procedure. Although this treatment may only be effective in a small group of patients, it is a new, straightforward procedure giving an excellent result in the face of a difficult problem.

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Mr G P J Mullan takes responsibility for the integrity of the content of the paper.
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