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CLINICAL RECORD

Balloon dilatation to treat plasmacytosis of the supraglottic larynx

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Abstract

Objective: We discuss the use of balloon dilatation to relieve supraglottic stenosis caused by mucous membrane plasmacytosis.

Case report: A 54-year-old man with a known diagnosis of mucous membrane plasmacytosis presented with dysphonia and worsening airway obstruction which required a tracheostomy. He underwent balloon dilatation of the supraglottic larynx using an angioplasty balloon within sequentially sized endotracheal tubes. This enabled successful decannulation, with minimal re-stenosis at eight-month follow up.

Conclusion: To our knowledge, this is the first reported case of supraglottic stenosis caused by plasmacytosis to be successfully treated using this method. We have shown that this minimally invasive technique deals effectively with a complex airway and minimises re-stenosis.

Key words: Larynx; Plasmacytosis; Supraglottis; Airway Obstruction; Balloon Dilatation

Introduction

Plasmacytosis is a rare, idiopathic condition that presents as soft tissue lesions involving the mucosa within the head and neck.[1] It commonly affects the oral cavity and oropharynx, but can also affect the larynx. There is only one case in the modern literature of plasmacytosis affecting solely the supraglottis.[2–4]

In this case study, we discuss the management of a patient with supraglottic stenosis due to plasmacytosis, using balloon dilatation. This is the first reported case to undergo this treatment. Balloon dilatation enabled decannulation with minimal re-stenosis at follow up.

Case study

A 54-year-old man had a 10-year history of oral plasmacytosis, which had been diagnosed and was being treated by the oral medicine team (with topical steroids and chlorhexidine mouthwashes). He presented to a local accident and emergency department with a two-day history of increasing airway obstruction and inspiratory stridor. He had also developed dysphagia to solids.

Prior to admission, the patient had been referred to the respiratory physicians with an episode of increased shortness of breath and 'wheeze', which had been treated as asthma with inhaled topical β2 stimulants and steroids, with minimal effect. Interestingly, in the last three years his oral plasmacytosis had become quiescent.

On initial assessment using a flexible nasendoscope, significant supraglottic stenosis and oedema were noted, with a number of thick mucosal bands extending between the posterior pharyngeal wall and the left arytenoid cartilage. Due to the severity of the stenosis, a surgical tracheostomy was performed to secure the airway.

The patient was subsequently referred to the ENT department at Salford Royal National Health Service (NHS) Foundation Trust, where the senior author had a specialised interest in complex upper airway management.

The patient underwent a diagnostic laryngo-tracheo-bronchoscopy under general anaesthesia. Examination revealed severe supraglottic stenosis particularly affecting the inter-arytenoid mucosa (Figure 1a). There was also a thick fibrotic band extending from the left arytenoid towards the lateral pharyngeal wall and obliterating the left pyriform fossa (Figure 2a). Suprastomal granulation was also present.

Initial surgical management consisted of resection of the pharyngeal band to re-establish a patent pyriform fossa (Figure 2b). The pharyngeal band was sent for histological analysis.

The supraglottic stenosis was then measured using sequentially sized, non-cuffed Portex endotracheal tubes (Portex, Smiths Medical, Watford, UK) introduced over a 4 mm, 0° Hopkins rod which allowed detailed visualisation of the larynx during the procedure.

A size 5.5 Portex endotracheal tube (outer diameter 8.0 mm) passed through the stenotic supraglottis with ease. This was gradually increased in 0.5 increments to a size 7.5 Portex endotracheal tube (outer diameter 10.4 mm), which was passed through the stenotic portion with gentle rotatory pressure, without causing mucosal trauma.

Once the endotracheal tube was correctly sited, a Boston Scientific angioplasty balloon catheter (12 mm; Boston Scientific, Natick, Massachusetts, USA) was inserted at a pre-measured length within the tube at the site of the stenosis and inflated to a pressure of 12 atmospheres for

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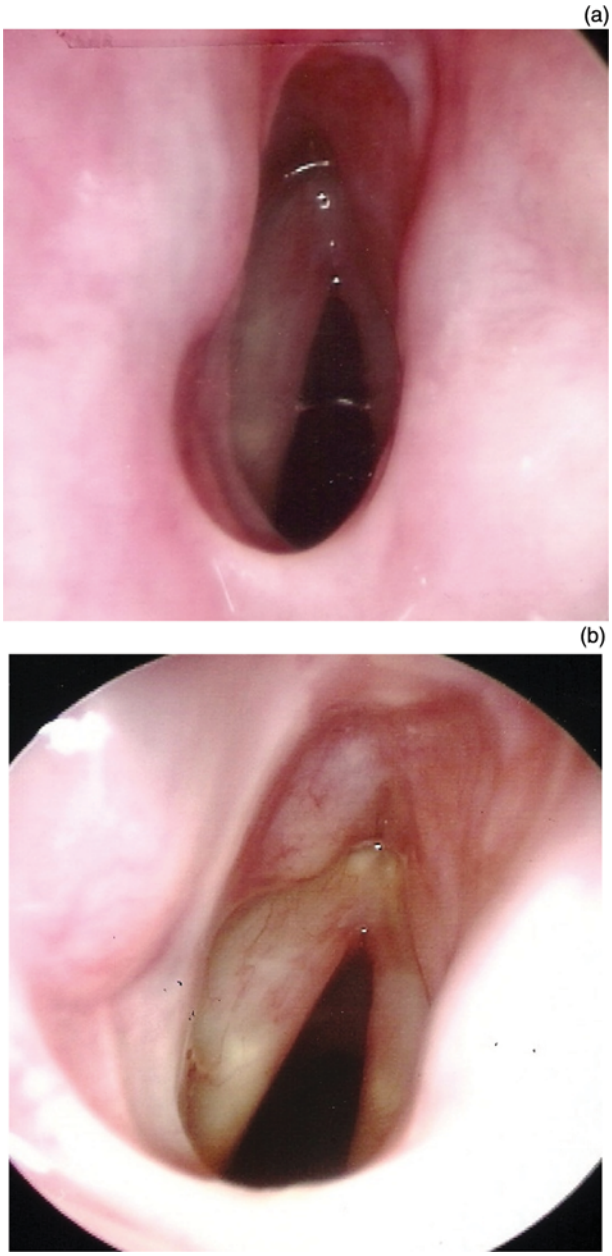

FIG. 1

Laryngo-tracheo-bronchoscopic views showing (a) significant supraglottic stenosis pre-operatively, and (b) improved airway with minimal re-stenosis eight months post-operatively.

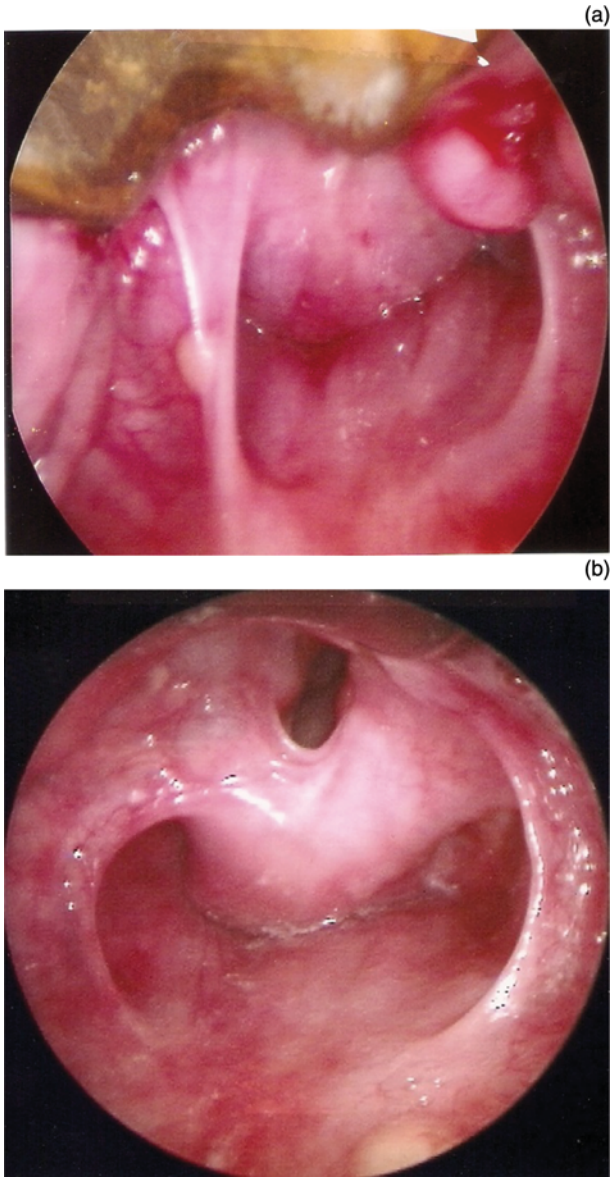

FIG. 2

Laryngo-tracheo-bronchoscopic views showing (a) a large mucosal band between the left posterior supraglottis and the left oropharyngeal wall, and (b) post-operative appearance following removal of the band.

approximately 2 minutes on 4 occasions (Figure 3). Following dilatation, no mucosal trauma was noted. At the end of the procedure, a Portex size 7.5 endotracheal tube passed with no resistance. Subsequent measurement of the circumference of the inflated balloon within the Portex size 7.5 endotracheal tube confirmed a dilated diameter of 12.5 mm (where diameter = circumference/π). A size 8 fenestrated Tracoe tracheostomy tube (Tracoe, Kapitex Healthcare, Wetherby, England) was left in situ.

Two weeks later, the patient was reviewed in the ENT outpatient department. Histological analysis of the surgical specimen was reported as showing chronic inflammatory cells with a high proportion of mature plasma cells (Figure 4). Immunostaining with cluster of differentiation 79a and 138 proteins further confirmed the presence of mature plasma cells. In addition, there was no evidence of kappa or lambda light chain restriction, which would have indicated the presence of myeloma or plasmacytoma. These findings increased the likelihood that plasmacytosis of the supraglottic larynx was the correct diagnosis.

The patient reported a marked improvement in his swallowing. Examination using fibre-optic nasendoscopy showed no recurrence of the fibrous bands within his pharynx, and a markedly improved airway.

Five months later, a second laryngo-tracheo-bronchosopic procedure was performed to remove a suprastomal granuloma by 'cold steel' dissection. Trial decannulation of the tracheostomy the following day was successful.

Eight months after dilatation, repeated microlaryngoscopy and endoscopic examination confirmed that the improvement in the supraglottic stenosis had been maintained (Figure 1b).

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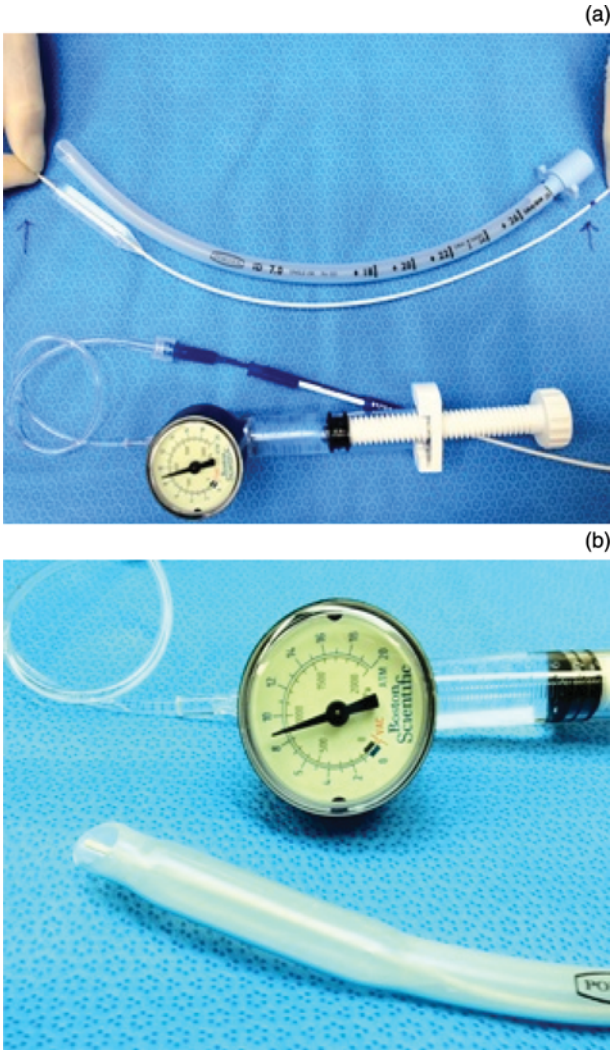

FIG. 3

Photographs demonstrating (a) pre-operative measurement of the required length of the angioplasty balloon, in order to accurately dilate the stenotic region, and (b) extent of balloon expansion within the endotracheal tube.

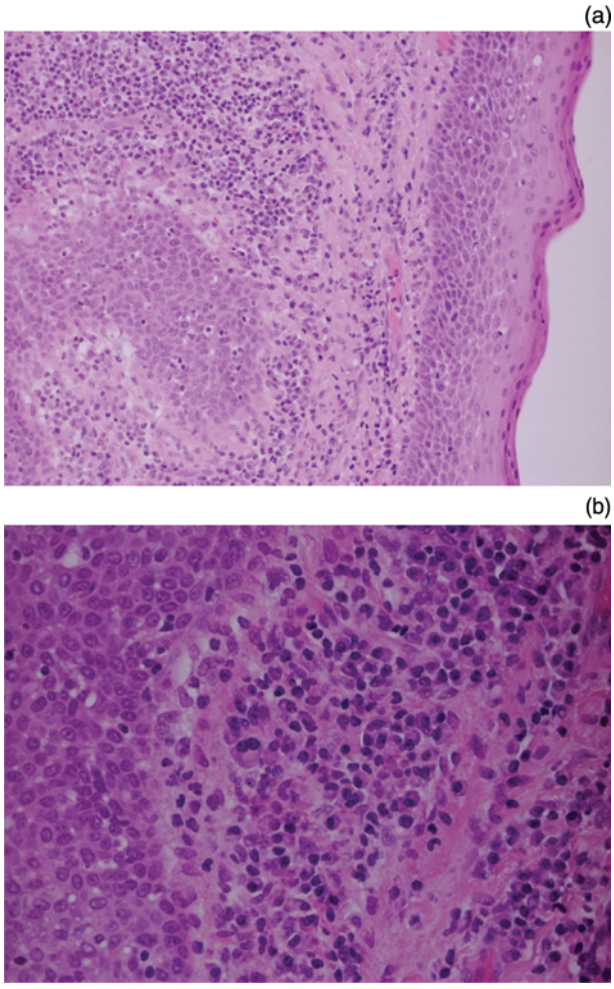

FIG. 4

Photomicrographs of pharyngeal mucosa viewed at (a) ×20 and (b) ×40, showing inflammatory cells with a large number of mature plasma cells. (H&E)

At the same time, a small residual tracheo-cutaneous fistula was formally closed.

Discussion

Mucous membrane plasmacytosis is a rare, benign condition first described by Zoon in 1952.[5] The aetiology and pathogenesis of the disease process are poorly understood. It is thought to affect both men and women after the fourth decade of life.[2]

In the oral cavity and oropharynx, plasmacytosis presents as well-circumscribed, soft, elevated lesions which cause painful, swollen lips, gums and throat. In the larynx, the lesions are classically described as having a cobblestone or warty appearance. These lesions can present with dysphonia and breathing difficulties, and generally involve the glottis and supraglottis; rarely, they involve the subglottis.

The histological features of plasmacytosis are described in Table I. As shown in Figure 4, the pharyngeal biopsy from our patient revealed an infiltrate of chronic inflammatory cells with a high proportion of mature plasma cells. Although immunostaining confirmed the presence of a high proportion of mature plasma cells, this tissue sample alone would not satisfy the criteria for histological diagnosis of plasmacytosis if the biopsy was considered in isolation. The final histopathological impression was given in the context of the clinical picture.

The treatment of plasmacytosis includes both medical and surgical approaches. However, due to limited literature on the subject, there is debate over the most effective treatment.

Medical care consists of topical and/or oral steroids along with additional adjuvant topical therapy (e.g. chlorhexidine mouthwashes), depending upon the site of involvement. Such regimens are proven to be effective in controlling lesions within the oral cavity.[3,6,7] Larger lesions causing

TABLE I
HISTOLOGICAL FEATURES OF PLASMACYTOSIS[1]

Large numbers of mature plasma cells
Mature plasma cells lacking pleomorphism
Russell bodies*
No κ or λ light chain restriction on immunostaining†

*Accumulations of immunoglobulin within plasma cell cytoplasm. †The presence of such light chain restriction would indicate myeloma or plasmacytoma.

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obstructive symptoms require surgical debulking using 'cold steel' dissection or laser resection. However, it has been shown that these patients often have progressive disease and worsening of symptoms on long-term follow up.[2] It has been postulated that this is due to an excessive inflammatory response causing fibrosis and consequent re-stenosis.

Ferreiro *et al.* have published a case series describing the management of upper aerodigestive tract plasmacytosis.[2] Their series consisted of nine patients, six of whom had manifestations within the supraglottis. These patients were managed by a combination of oral steroids, topical steroids, laser and surgical resection. It was noted that oral steroids improved symptoms and airway obstruction in the short term. However, two cases required tracheostomy for worsening airway obstruction due to disease progression. Unlike our patient, neither of these two patients were able to be decannulated.[2]

- **Plasmacytosis can affect the upper aerodigestive tract**
- **Laryngeal involvement can cause dysphonia and airway obstruction**
- **Due to limited publications, the best management is unknown**
- **Previously reported surgery failed to prevent re-stenosis and disease progression**
- **In this case, balloon dilatation improved the airway and minimised re-stenosis**

In cases such as our patient's, we feel it is important to consider a minimally invasive technique in an attempt to limit further mucosal fibrosis.

In our patient, we used the novel technique of inflating an angioplasty balloon within an endotracheal tube, an approach previously reported by the senior author.[8] The controlled, fusiform expansion of the endotracheal tube protects the mucosa against the sudden, explosive force of inflating an angioplasty balloon. This method of dilatation prevents the shearing forces on the mucosa that accompany linear dilatation. An alternative dilatation technique uses sequential rigid dilators. The major disadvantage of this technique is the mucosa trauma sustained at the site of maximal stenosis, due to the application of longitudinal shearing forces at the point of maximal resistance. We believe our technique reduces the risk of significant mucosal damage, with subsequent fibrosis and re-stenosis, and achieves our aim of avoiding healing by secondary intention. After undergoing this technique, our patient was found to have minimal re-stenosis at eight months post-operatively.

Conclusion

We believe this to be the first published case in which balloon dilatation within an endotracheal tube was used to improve the supraglottic airway in a patient with mucosal plasmacytosis affecting the larynx. We have shown that this minimally invasive technique deals effectively with a complex airway and minimises re-stenosis (as observed eight months post-operatively).

Acknowledgement

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Mr S G Mistry takes responsibility for the integrity of the content of the paper
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