

Balloon Dilatation of the Trachea as Treatment for Idiopathic Laryngotracheal Stenosis

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The treatment of choice for idiopathic laryngotracheal stenosis is tracheal resection and anastomosis, although some authors prefer more conservative management.

Between January 1, 1996 and January 1, 2005, 8 patients—all women—with idiopathic laryngotracheal stenosis were treated in the chest surgery department of the Hospital Universitario La Fe in Valencia, Spain. One case was treated by means of surgery and so was excluded from this study. The remaining 7 women were treated by tracheal balloon dilatation; 4 required just 1 dilatation (and remained asymptomatic), 2 required 2 dilatations, and 1 required 4 dilatations. The median symptom-free interval was 25.5 months, and there was no associated mortality or morbidity.

We conclude that balloon dilatation, which was not associated with mortality or morbidity, is a suitable treatment option for idiopathic laryngotracheal stenosis.

Key words: *Idiopathic laryngotracheal stenosis. Balloon dilatation. Tracheal resection.*

Introduction

Idiopathic laryngotracheal stenosis (ILS) is a rare disease, the cause of which is unknown. Typically occurring in women aged 20 to 60 years old, it is characterized by nonspecific inflammation of the upper third of the tracheal submucosa or the subglottic region and may develop into circumferential cicatricial stenosis. Patients typically experience mild dyspnea lasting for months or years before progressing to dyspnea at rest, wheezing, or stridor. As a result, many patients may be incorrectly diagnosed with and treated for asthma.¹ As far as ILS diagnosis is concerned, a patient should not have any history of upper airway stenosis.²

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Dilatación traqueal neumática en el tratamiento de la estenosis traqueal idiopática

El tratamiento de elección de la estenosis traqueal idiopática es la resección-anastomosis traqueal, aunque algunos autores defienden el tratamiento conservador.

Entre el 1 de enero de 1996 y el 1 de enero de 2005, en el Servicio de Cirugía Torácica del Hospital Universitario La Fe de Valencia se trató a 8 pacientes con estenosis traqueal idiopática, en un caso mediante cirugía, por lo que se excluyó del estudio. Todos los pacientes eran mujeres y 7 de ellas fueron tratadas mediante dilataciones traqueales neumáticas periódicas. Únicamente han precisado una dilatación 4 pacientes, que hasta ahora permanecen asintomáticas. Dos han necesitado una segunda dilatación y una paciente ha requerido 4. La mediana de intervalo sin síntomas fue de 25,5 meses. No hubo mortalidad ni morbilidad asociada.

En conclusión, la dilatación traqueal neumática es una opción terapéutica adecuada en el tratamiento de la estenosis traqueal idiopática, sin morbilidad atribuible a la técnica.

Palabras clave: *Estenosis traqueal idiopática. Dilatación traqueal neumática. Resección traqueal.*

The treatment of choice is tracheal resection and anastomosis.²⁻⁴ That said, a number of authors defend a more conservative treatment for reasons such as the possibility of lesion recurrence, the morbidity and mortality associated with surgery, and the high percentage of patients requiring palliative treatment after resection.⁵

Our aim was to evaluate results for a series of patients with ILS who received conservative treatment in the form of balloon dilatation of the trachea.

Case Description

Between January 1, 1996 and January 1, 2005, 103 patients with laryngotracheal stenosis—idiopathic in 8 cases—were treated in the chest surgery department of the Hospital Universitario La Fe in Valencia, Spain. One patient who received surgical treatment was excluded from the study, leaving 7 patients who received balloon dilatation treatment on a regular basis. Patients were followed up for at least 21 months.

The following medical data for the patients were analyzed: sex and age; medical history; date of diagnosis; fiberoptic bronchoscopy; tracheal computed tomography; stenosis type, length, and internal lumen; number of procedures; and disease-free interval. Possible causes of upper airway stenosis were excluded. Under general anesthesia all the patients underwent the same procedure, as follows: a fiberoptic bronchoscope was introduced through a laryngeal mask and balloon dilatation of the trachea was performed.⁶

All the patients in the series were women. Mean age (SD) at the time of diagnosis was 59.14 (12.49) years (range, 42-77 years). All the patients had progressive dyspnea. The lesion was located in the upper third of the trachea in all 7 of the patients, and there was subglottic involvement in 5 of the patients. All the lesions were circumferential, with a mean (SD) length of 1.74 (0.42) cm (range, 1.0-2.2 cm) and an internal lumen of 7.55 (1.61) mm (range, 5.0-10.0 mm). Of the 7 patients, 4 required just 1 dilatation and are still asymptomatic (symptom-free interval range, 21-96 months); 2 patients required a second dilatation at 53 and 60 months, respectively, and were asymptomatic after 8 and 32 months, respectively; and finally, 1 patient required a total of 4 dilatations (at 27, 10, 12, and 52 months), and is still asymptomatic (Table). The median symptom-free interval was 25.5 months.

There was no mortality or morbidity associated with the series. One patient died 21 months after dilatation as a consequence of liver cancer; this patient had undergone surgery 9 months after the dilatation and had had no complications as a consequence of the orotracheal intubation.

Discussion

ILS is a rare entity that is diagnosed by ruling out known causes of subglottic stenosis, such as postintubation tracheal stenosis, tracheal trauma (both external and internal, and including inhalation and radiation burns), specific and nonspecific tracheal infections (bacterial tracheitis, tuberculosis, histoplasmosis, and diphtheria), collagen disorders and vasculitis (Wegener granulomatosis, recurrent polycondritis, polyarteritis, scleroderma, and sarcoidosis), amyloidosis, and finally, congenital causes.³

Although authors such as Walner et al⁷ and Koufman⁸ have linked the presence of gastroesophageal reflux to the

appearance of laryngeal lesions, only 1 of the patients in our series had a history of such lesions. Nonetheless, we do not believe that reflux was the cause of this patient's symptoms. If this were the case, we would have observed inflammation of the vocal cords, yet no inflammation was evident in the diagnostic or therapeutic fiberoptic bronchoscopy procedures performed. Furthermore, reflux is equally prevalent in men and women, whereas ILS occurs exclusively in women.

A number of treatments are available for ILS, ranging from more conservative therapies to surgery. As far as surgery is concerned, the study of reference was published in 1993 by Grillo et al,² who described the procedure that is currently the surgery of choice for this condition, namely, single-staged laryngotracheal resection. This procedure produced good or excellent results in 91% of the 35 patients in their study. Ashiku et al³ published a review and update for the same study, and they likewise reported good or excellent results for 91% of the series of 73 patients. Dedo and Catten,⁵ on the other hand, were of the opinion that, since ILS is a progressive disease, the best therapeutic option consists of regular palliative treatment. The authors arrived at this conclusion as a consequence of the poor outcomes they obtained for resection: stenosis reoccurred in all 7 of the patients undergoing resection. Of the 50 patients treated by Dedo and Catten, 13 required tracheostomy, 16 required more than 10 procedures each, and 21 were still disease-free after a fairly lengthy period. The median number of procedures per patient was 6 (mean, 8).

These data would lead us to conclude that the natural history of ILS is not well understood and that its course is unpredictable. Noteworthy conservative treatments include tracheal dilatation,^{6,9} local corticosteroid injection, or laser treatment. It is clear that aggressive and repeated treatments such as tracheostomies, stenting, and dilatation using rigid bronchoscopes merely contribute to enlarging the affected tracheal area, and, furthermore, complicate any possible surgical repair.^{3,6}

In the light of our own results, we agree with Park et al⁴ in that patients with ILS can be divided into 2 groups: patients with intermediate detectable ILS, requiring just 1 or 2 dilatations, and those with more complex, recurrent forms of the disease who require resection. We conclude, therefore, that balloon dilatation, which has no associated mortality or morbidity, is a suitable treatment option for ILS.

Clinical Characteristics of Patients With Idiopathic Laryngotracheal Stenosis

Code	Age, y	Sex	Presentation	Date Diagnosed	Subglottic Involvement	Dilatations	Time Elapsed, m*	Current Situation
1	48	Female	Progressive dyspnea	11/27/2003	No	1	24	Asymptomatic
2	77	Female	Progressive dyspnea	12/18/2002	Yes	1	24	Asymptomatic
3	72	Female	Progressive dyspnea	07/11/2001	Yes	1	21	Deceased (liver cancer)
4	62	Female	Progressive dyspnea	11/01/1996	Yes	1	96	Asymptomatic
5	54	Female	Progressive dyspnea	08/10/1998	No	2	53 and 32	Mild stridor
6	59	Female	Progressive dyspnea	01/27/2000	Yes	2	60 and 8	Asymptomatic
7	42	Female	Progressive dyspnea	10/24/1997	Yes	4	27, 10, 12, and 50	Asymptomatic

*Time elapsed between tracheal dilatations.

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