

# Contemporary Management of Idiopathic Laryngotracheal Stenosis



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## KEYWORDS

- Pearson procedure • Idiopathic laryngotracheal stenosis • Subglottic tracheal resection
- Airway reconstruction

## KEY POINTS

- Idiopathic subglottic stenosis is a rare diagnosis affecting predominantly women, whose etiology is yet to be understood.
- Endoscopic treatment can be successful for some patients with idiopathic subglottic stenosis, yet recurrence rates are high despite multimodal therapy.
- Subglottic tracheal resection with synchronous laryngeal resection and preservation of the recurrent laryngeal nerves can provide excellent and durable outcomes for patients with idiopathic subglottic stenosis.

## INTRODUCTION

Idiopathic subglottic stenosis (ISS) was first described in the 1970s,<sup>1</sup> and is a rare but well-described indication for airway intervention and subglottic tracheal resection. As described, ISS is a diagnosis of exclusion, made after ruling out other common causes of stenosis (**Box 1**).

Controversy exists regarding the optimal treatment for ISS; thus, it is imperative that the diagnosis is correct and all other etiologies are ruled out by history, blood work (eg, antineutrophilic cytoplasmic antibody for granulomatosis with polyangiitis, angiotensin-converting enzyme for sarcoidosis) and imaging. These patients are typically women aged 20 to 60 years, with a typical history of insidious onset dyspnea and noisy breathing that often lasts months to years before diagnosis.<sup>2,3</sup> Often, these patients are treated for other conditions or misdiagnosed as having “asthma” before the diagnosis of ISS. Although

most of these patients generally do not have a history of prolonged intubation, many have a history of a short intubation for a surgical procedure.<sup>3-6</sup> There are no data to support that short periods of intubation (such as for elective surgical procedures) contribute to the development of the stenosis. As an anecdotal observation, short stature women who develop postintubation subglottic stenosis likely experience trauma to the subglottic airway because they have a smaller trachea; the clinical tendency is to use a standard sized endotracheal tube such as a size 7.0 or 7.5 for routine intubation, which is actually too large for these patient. Also, gastroesophageal reflux disease has been postulated as a potential causative or contributing factor in the development of ISS. There are no definitive data supporting this suggestion, however, and many patients presenting with ISS have no history of gastroesophageal reflux disease, although it may contribute to the development of ISS in some patients.<sup>6-8</sup> Because

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**Box 1****Causes of subglottic tracheal stenosis**

Postintubation

Trauma

Inhalational burns

Radiation

Collagen vascular diseases (including granulomatosis with polyangiitis, sarcoidosis, amyloidosis, relapsing polychondritis, etc)

Infectious (eg, tuberculosis)

Tracheomalacia

more than 90% of reported patients with ISS are women, a hormonal influence has also been suggested. Estrogen, in particular, has been suggested as causative because it has been shown to play a role in fibrosis and keloid formation, but its role in ISS is yet to be elucidated.<sup>6,9</sup> Familial predisposition has not been established, although case reports of sisters and mothers and daughters developing ISS have been reported.<sup>10</sup> Some series have also found that body mass index is increased in women with ISS.<sup>11</sup>

ISS is defined by the endoscopic findings, which include a circumferential cicatricial stenosis that develops secondary to nonspecific inflammation, affects the proximal trachea and subglottic area, and is typically less than 2 cm in length.<sup>2,12</sup> Compared with patients with tracheal stenosis caused by other conditions, ISS tends to be shorter and located more proximally in the trachea.<sup>3</sup> Stenoses can be classified based on the Myer-Cotton classification, which equates endotracheal luminal size with degree of airway stenosis (**Table 1**).<sup>13</sup> Biopsy of the lesions shows fibrosis with or without acute inflammation.<sup>14</sup>

### THERAPEUTIC OPTIONS: ENDOSCOPIC TREATMENT

Debate exists in the literature regarding the optimal treatment modality for patients with ISS. Generally, these patients are first managed endoscopically, both for diagnostic and therapeutic

purposes. A first attempt at endoscopic treatment is often tried in the majority of patients with variable success.<sup>6,15</sup> Also, because the pathophysiology of ISS remains under debate, some investigators feel that endoscopic therapy is most appropriate because this process is slowly progressive and may recur in the future, even after definitive surgical resection.<sup>16</sup> Performing an endoscopic dilation of a subglottic stenosis can be therapeutic, yet can also aid in treatment planning, because it allows the surgeon to determine the length and severity of the stenosis. Advantages of endoscopic therapy include the lack of skin incision and the ability to discharge most patients on the day of the procedure. Also, endoscopic therapy is ideal for patients with ongoing active inflammation because these patients are not considered to be good surgical candidates until the active inflammation resolves.<sup>3</sup> Patients with long (>2 cm) or complex stenoses (with or without a malacic component) are at greater risk for failure of endoscopic therapy and thus considered to be better candidates for surgical management.<sup>3,14</sup>

### Endoscopic Dilations

Dilation can be done using flexible bronchoscopy with balloon dilation, or rigid bronchoscopy with or without the use of gum-tipped bougie dilators. Performing dilation alone has been found to have a high rate of failure in patients with subglottic stenosis owing to all causes; thus, adjunctive modalities have been sought to improve these results.<sup>17</sup> Although patients with a diagnosis of ISS had a longer interval between dilations compared with other diagnoses, patients still required an average of 3 dilations, with some patients requiring up to 24 procedures.<sup>18</sup> Halmos and colleagues<sup>19</sup> found that patients with ISS required the most dilation bronchoscopies compared with other diagnoses. In contrast, another series found that, compared with patients who underwent dilation for stenosis caused by other etiologies, patients with ISS were found to have a significantly longer time between first and subsequent dilation.<sup>20</sup> Voice quality and production is a concern for many patients with ISS as because the stenosis is quite proximal and treatment can impact the vocal cord anatomy. The use of balloon dilation has been shown in a small series to improve dysphonia, which may be an advantage of endoscopic therapy compared with open surgical management.<sup>21</sup> Injections of the stricture with intralesional steroid or mitomycin have been described.<sup>22</sup> Although data are lacking for the efficacy of intralesional steroid injection to prevent stenosis recurrence, a small series found that the average interdilation interval was

**Table 1**  
**Myer-Cotton grade of airway obstruction**

Grade	Obstruction (%)
I	50
II	51–75
III	>70 with any detectable lumen
IV	No detectable lumen

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