Cross-sectional Study

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Laryngotracheal stenosis: a single-center retrospective analysis of endoscopic treatment strategies and recurrence

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Background: Laryngotracheal stenosis is a process of fibrosis that results in airway obstruction; it may be congenital or acquired. Acquired cases are due to iatrogenic, traumatic, infectious and autoimmune causes. Patients present with a spectrum of breathing difficulties that might be fatal.

Objectives: This article presents a unique retrospective cross-sectional study of patients with laryngotracheal stenosis who had endoscopic dilatation, and some had open surgical procedures to describe the evolution after the therapy, to compare the outcomes of the dilatation in comparison to surgical interventions and associated complications in other studies, to provide knowledge to help in dealing with these patients, and to enable educated, independent patient decision-making.

Material and methods: In this observational and descriptive study, we aimed to highlight the clinical features, management, and treatment outcomes among twenty-nine patients with laryngotracheal stenosis who were managed by open and endoscopic surgical intervention over a period of 5 years. Data were collected retrospectively from the patients' medical records from February 2016 until July 2022 at a hospital in Jerusalem, which is a tertiary healthcare facility and the only referral center for similar cases from the West Bank, East Jerusalem, and Gaza, with a population of around 8 million. Tables and graphs are used to highlight the statistical study's findings. Data were analyzed using Microsoft Excel software.

Results: Twenty-nine patients were involved in the study, with an average age of 32.2 years; 51.7% of them were males. The patients had one or more chronic conditions, such as hypertension and coronary artery disease. The majority of patients (65.5%) had stenosis as a result of orotracheal intubation, and the most common site was the subglottic (68.9%). According to the Cotton–Myer classification, 44.8% of the patients were classified in grade III, 34.4% were included in grade I, 13.7% in grade II, and 6.8% in grade IV. Six patients (20.68%) received surgery for stenosis, and 23 patients had an endo-laryngeal dilatation. Restenosis happened in (58.6%). Mortality rates are almost negligible.

Conclusion: Subglottic stenosis is still a challenging condition to manage. The authors describe a single-center experience approach dealing with these conditions. Long-term follow-up for these cases is mandatory as the recurrence rate is still high.

Keywords: endoscopic ballon dilation, endoscopy, laryngotracheal stenosis, subglottic stenosis, tracheostomy

Introduction

Laryngotracheal stenosis is one of the most challenging topics in thoracic surgery, as it has multiple etiologies, no definite treatment, and a high recurrence rate. The main pathophysiology is believed to be abnormal wound healing that leads to hypertrophic scar formation and narrowing of the airway[1].

The most common cause of laryngotracheal stenosis is iatrogenic post-intubation[2]. Many factors can increase the risk of stenosis after intubation, including those relevant to the patient, such as body mass index, the presence of other comorbidities, and a previous history of intubation for more than 48 h[3]. Other factors are treatment-related, like the prolonged period of intubation and the large size of the tube[2].

HIGHLIGHTS

- Laryngotracheal stenosis has multiple etiologies, with no definite treatment and with a high recurrence rate.
- The Myer–Cotton system is a grading system used for classifying central airway stenosis as a percentage for the reduction of the cross-sectional area of the obstructed airway.
- The most common postoperative complications after laryngotracheal stenosis are granulations, restenosis and dysphonia.
- The most common cause for laryngotracheal stenosis is iatrogenic post-intubation.

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Laryngotracheal stenosis can be caused by various causes such as congenital, trauma, infectious, radiation, chemical inhalation, thermal injuries,[1] rheumatological and autoimmune diseases, and is most commonly reported in granulomatous polyangiitis (GPA)[2].

Idiopathic laryngotracheal stenosis is considered a diagnosis of exclusion and is reported mainly in females with a median age of 47 years old[4]. The pathophysiology of this subtype is the same as previously mentioned process of fibrosis without any earlier trauma or injury[1].

The clinical presentation of laryngotracheal stenosis includes shortness of breath, wheezing, and stridor. Biphasic stridor happens with subglottis stenosis, while inspiratory stridor with supraglottis stenosis. On the other hand, tracheal stenosis presents with an expiratory stridor[4].

Evaluation for patients suspected of having laryngotracheal stenosis begins with a thorough history and physical examination. A flexible fiberoptic laryngoscope helps to identify any upper airway problems above the vocal cord. However, rigid direct laryngoscopy and flexible fiberoptic bronchoscopy are the gold standards for subglottis evaluation, as they have both diagnostic and therapeutic benefits[5].

Dilatation can be done with flexible bronchoscopy using balloon dilators or rigid bronchoscopy with or without bougie dilators. To maximize the benefit, adjuvant therapy can be used as an intralesional steroid or mitomycin c injection during dilatation[6]. Small groups in the previous literature found that intralesional steroid injection increased the interdilation interval[2].

On the other hand, there are multiple surgical approaches that can be done in more severe cases, and that depends on the level and extent of the stenosis, with either resection and anastomosis, laryngotracheal reconstruction, or cervical side tracheoplasty[5].

Material and methods

This retrospective cross-sectional study was conducted at a Hospital in Jerusalem, which is considered to be the regional referral center for tracheal and thoracic cases in the Palestinian Territories, serving approximately eight million people across the West Bank, East Jerusalem, and Gaza Strip. Patient data were retrospectively collected from medical records, encompassing follow-up visits, surgical procedures, and interventions performed from February 2016 to July 2022. A cohort of 29 patients diagnosed with laryngotracheal stenosis was included. Analyzed variables included age, gender, comorbidities, etiology of stenosis, sub-sites of stenosis, Cotton–Myer classification, surgical approaches, patient outcomes, and complications. Data were processed using Microsoft Excel and visualized through graphs and tables, with a comparative analysis of existing literature. Patients included in our study had a history of prolonged (24 h) or traumatic intubation, obvious temporal relation to uncomplicated brief intubation, neoplasm or malignancy, as well as autoimmune, vasculitis, radiation, traumatic, or iatrogenic causes. The stenosis had to involve the subglottis, glottic, supraglottic, and tracheal stenosis. Some patients who underwent operative procedures before treatment at our institution were also included. Pediatric patients with congenital causes were excluded due to the differences in etiology and treatment approaches. This work has been reported in line with the STROCSS criteria[7].

Surgical techniques

In our study, the majority of patients, twenty-three (79.3%), were managed with endoscopic laryngeal dilatation. We will describe our technique for endoscopic tracheal dilatation. The open surgical approach will not be discussed in this article, as it falls outside the scope of our study. For balloon dilatation, flexible bronchoscopy was utilized. The patient was placed in a supine position, with eyes shielded to prevent contamination from bronchial secretions and arms flexed. An appropriate-sized laryngeal mask airway (LMA) was fitted with an L-shaped adaptor to facilitate bronchoscope insertion while maintaining ventilation, a direct laryngoscope was used for high laryngotracheal stenosis (Fig. 1).

The procedure entailed advancing the bronchoscope to and beyond the stenosis site, clearing secretions, and subsequently inserting a dilating balloon through the bronchoscope's working channel. Balloon size selection was contingent upon the stenosis diameter, with sizes typically exceeding 10 mm if the stenosis could be traversed with the bronchoscope. During inflation, sedation levels were increased to ensure patient safety, with continuous monitoring of oxygen saturation to maintain levels above 90%. The procedure involved staged dilatation with varying balloon sizes, with careful monitoring to prevent over-inflation and to assess for complications like perforation or bleeding. Post-procedure, patients received systemic and inhaled steroids to mitigate inflammation. (Fig. 2).

In case the bronchoscope could not bypass the stenosis, a guidewire was placed under fluoroscopic guidance, followed by

Figure 1. Direct laryngoscopy.

Figure 2. Using a 5 mm balloon size without camera use for distal navigation through direct laryngoscopy.

balloon advancement and dilatation as previously described. Patients underwent follow-up bronchoscopy 2 weeks post-procedure to assess dilatation response, with further management tailored based on the degree of stricture improvement (Fig. 3A and B). A multidisciplinary team approach was adopted for cases resistant to balloon dilatation, exploring alternative interventions such as tracheal resection, permanent tracheostomy, stenting, or T-tube insertion.

Results

This study identified records from twenty-nine patient records. Ranging in age from one month to 60 years, with a mean age of 32.2 years. The cohort included 15 males (51.7%) and 14 females (48.3%). Comorbidities were absent in 18 patients (62.1%), while 11 (37.9%) presented with one or more (Table 1). Prolonged orotracheal intubation (pOTI), which is defined as lasting more than 7 days, was the predominant cause of stenosis in 18 cases (62%) (Table 2) (Fig. 4). Subglottic stenosis was the most frequent site that was found in 20 patients (69%), followed by tracheal stenosis in 9 cases (31%). According to the Cotton–Myer classification, the distribution was: grade I (34.4%), grade II (13.7%), grade III (44.8%), and grade IV (6.9%) (Table 3) (Fig. 5). Complications included restenosis in 17 cases (58.6%), with no instances of complete obstruction, and no mortality was reported during the follow-up period of 1 year. Six patients (20.6%) had open surgery due to failed endoscopic dilation. These patients underwent tracheoplasty, tracheolaryngeal reconstruction, and tracheal resection. Thirteen patients (44.8%) underwent tracheostomies.

Discussion

Based on our findings, the average age in our study group was 32.2 years. This is lower than the average age reported by del Carmen *et al.*[8] of 42.6 years and by Hseu *et al.*[9] of 48 years. The distribution of male 51.7% and female 48.2% was found to be similar to that reported by del Carmen *et al.*[8] (57.5% and 42.2%, respectively).

Regarding the associated comorbidities, in 12 patients (41.3%), one or more comorbidities were present, lower than that reported by del Carmen *et al.*[8] (64%); and higher than that reported by Reyes Solarte AJ (25%)[10].

The most common presenting symptoms for all the patients in our study were shortness of breath and stridor. Other commonly reported symptoms are coughing and changes in voice, as reported by Nikola *et al.*[11]

In our study, the most common site of stenosis is the subglottis, reported in 20 cases (69.9%), which is higher than that reported by Irene del Carmen (18.18%)[8] and also higher than what was reported by Nikola *et al.*[11] (33%), followed by tracheal stenosis,

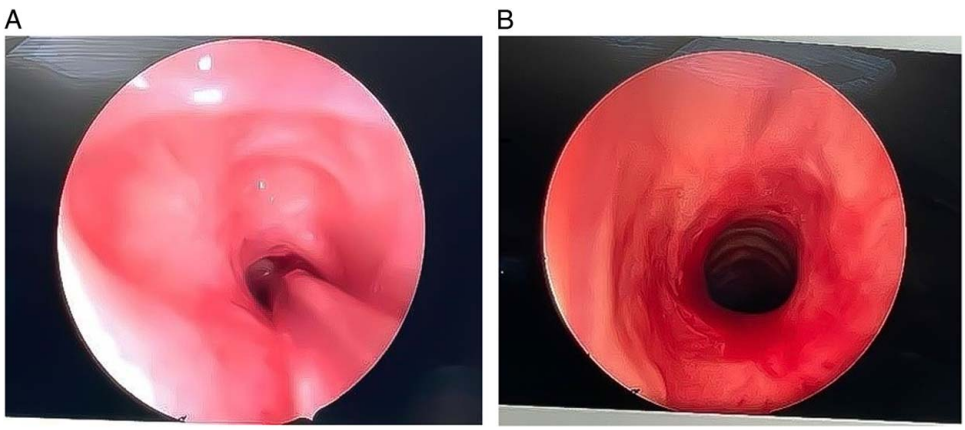

Figure 3. (A) A patient with grade III laryngeotreacheal stenosis. (B) A patient with grade III laryngotracheal stenosis after endoscopic dilatation.

Table 1

Comorbidities in patients in our study

Comorbidity[a]	Number	(%)
Hypertension	4	13.7
Diabetes mellitus type 1 and 2	3	10.3
Thyroid disorders	3	10.3
Heart disease	3	10.3
Wegener 's disease	1	3.4

[a]Some patients had more than one comorbidity.

Table 2

Etiology of laryngotracheal stenosis of the patients included in our group

Etiology	Number	(%)
Post-intubation	18	62
Idiopathic	8	27.5
Trauma	2	6.8

which is reported in 9 cases (31%), which is lower than that reported by Irene del Carmen (42.42%)[8], and also lower than what was reported by Nikola *et al.*[11] (44%).

Considering the causes of stenosis, the most common cause reported in our study is post-intubation, found in 18 cases (62.0%), higher than that reported by Hseu *et al.*[9] (25%). Eight cases (27.5%) were reported to be idiopathic, higher than that reported by Irene del Carmen (3.03%)[8]. Regarding the analysis of patients who developed stenosis post-intubation, 15 patients (83.3%) were intubated for more than 7 days, and 3 patients (16.6%) were intubated for less than 7 days, which is almost similar to what was reported by Irene del Carmen (78.26%) and (13.04%), respectively[8]. In a study by Nair *et al.*[12] patients who developed stenosis post-intubation were intubated for a period of 7–14 days, with a mean of 8.75 days.

The Myer–Cotton system is a grading system used for classifying central airway stenosis. The endotracheal tube is used to determine the reduction in cross-sectional area of the obstructed airway. It classifies the degree of stenosis into four groups. When stenosis is less than 50%, it is grade 1, when it is from 51 to 70%, it is grade 2, when it is from 71 to 99%, it is grade 3 and when no detectable lumen is observed, it is grade 4[13].

According to Cotton–Myer's classification, grade 1 was reported in 34.4% of cases, grade 2 in 13.7% of cases, grade 3 in 44.8%, and 6.8% of cases were reported as grade 4. These results are similar to those reported by Irene del Carmen; 27.2% as grade 1, 18.1% as grade 2, 51.5% as grade 3, and 3% as grade 4[8]. In a study by Nikola *et al.*[11] 38% of patients were classified as grade 1, 13% as grade 2, 27% as grade 3, and 19% as grade 4.

The most common postoperative complications are granulations, restenosis, and dysphonia[11]. Restenosis is the main complication found in our study in 17 cases (58.6%), which is lower than that reported by Irene del Carmen (67.5%)[8], and it is higher than what was reported by Nikola *et al.*[11] as (19%).

In our study group, 6 patients (20.68%) underwent open surgery and 23 patients (79.3%) underwent pneumatic dilatations by endo-laryngeal route, which is similar to what was reported by Nikola *et al.*[11] (20%) underwent open surgery and (80%) were treated with balloon dilatations.

Between the patients who underwent repeated pneumatic dilations by endo-laryngeal route, the mean time interval between procedures was 5.53 months, which is lower than that reported by Hseu *et al.*[9], which was 13.7 months. The average number of dilatation sessions was 3.4, which is higher than what was reported by Vorasubin *et al.*[14], which was 1.5.

The main aim of subglottic stenosis treatment is to maintain airway patency and minimize dyspnea, other objectives include maintaining the voice and swallowing. However, the study applies the prevention and decannulation of a tracheotomy as more objective end criteria to assess therapy efficacy[2].

Endoscopic approaches are recommended to prevent open-neck surgical morbidity; they have been documented in many different variants. This usually includes deciding whether or not to inject steroids and the dilation technique, commonly in conjunction with CO2 laser (Fig. 6) scar lysis or radial incisions[9,15].

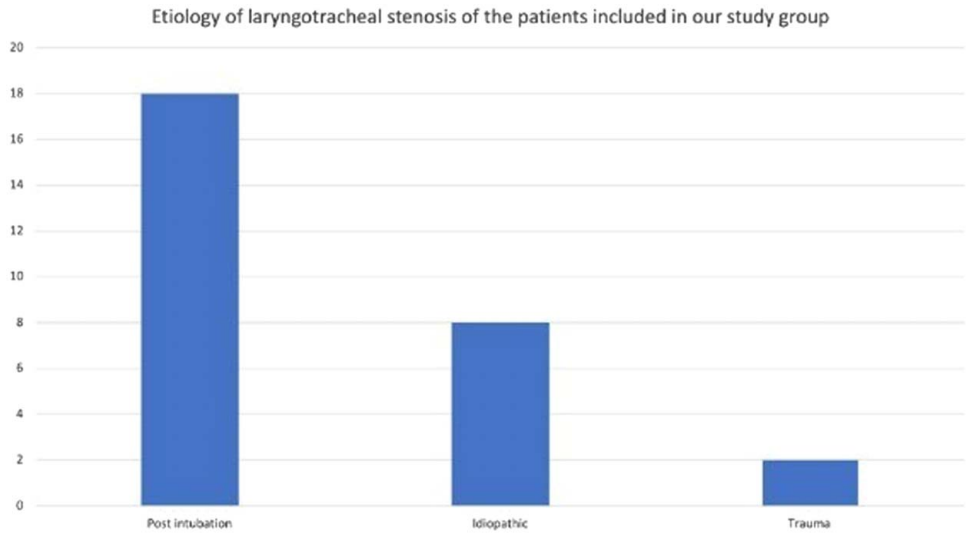

Figure 4. Etiology of laryngotracheal stenosis of the patients included in our study group.

Table 3

Degree of laryngotracheal stenosis, according to the Cotton–Meyer classification

Degree of stenosis, according to Cotton–Myer classification	Number	(%)
I	10	34.4
II	4	13.7
III	13	44.8
IV	2	6.8
Total	29	100

In addition to being less invasive for the patients, endoscopic balloon dilatation was preferred over open surgery because it guarantees the highest voice quality with the least invasive technique. However, long-term follow-up is recommended due to the high risk of recurrence and the requirements for revision surgery[15].

On the other hand, the choice between the different open surgery techniques resection and anastomosis or reconstruction with or without grafting depends on the degree and extent of the stenosis[2]. Patients with moderate to severe subglottic stenosis or multilevel airway obstruction are more likely to benefit from open surgical procedures, although they have a higher risk of morbidity[5]. When compared to cricotracheal resection (CTR), endoscopic dilatation and open reconstructive laryngotracheoplasty have been associated with better voice outcomes[16].

Even though tracheostomy preserves the patency of the airway, it has an impact on breathing, swallowing, voice, and quality of life[2]. Only those with severe subglottic stenosis, failed first surgery, additional airway obstructions, and/or chronic comorbidities are often tracheostomy-dependent[5].

This study is limited by its retrospective observational nature with a small population. It was challenging to analyze the data and compare it to other studies since some old medical records lacked important data we needed, there was no objective way to evaluate the degree of the stenosis, and it was hard to bring Gaza's patients back to follow-up due to Israeli occupation restrictions.

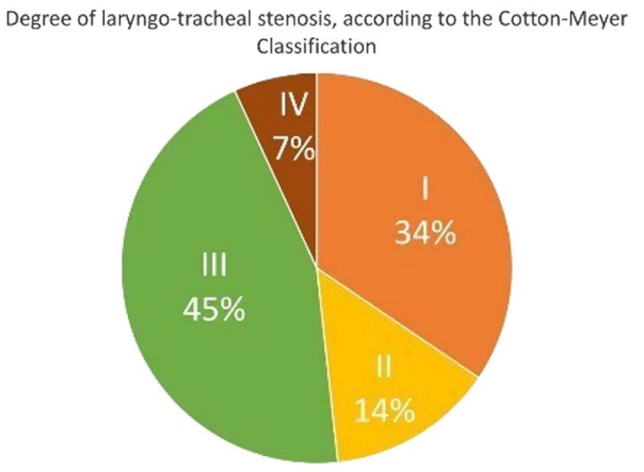

Figure 5. Degree of laryngotracheal stenosis, according to the Cotton–Meyer classification.

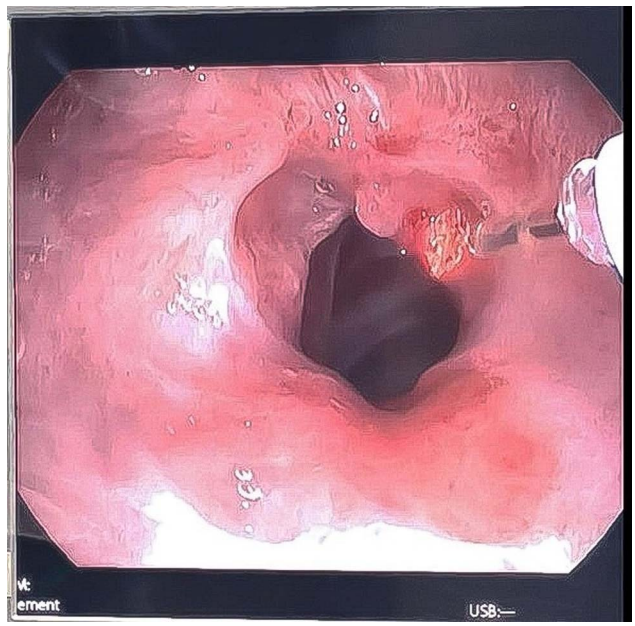

Figure 6. Using Co2 laser on a well- formed granulation tissue as an adjunct with endoscopic dilatation.

Conclusions

The study primarily used endoscopic balloon dilatation for most cases, but due to recurrence and revision surgery, long-term follow-up is necessary. Only a small proportion needed open procedures, and severe subglottic stenosis required a tracheostomy. Although the mortality rate is lower than previous reports, a multidisciplinary approach is recommended for treating subglottic stenosis.

Ethical approval

The studies were conducted in accordance with the local legislation and institutional requirements. An ethical approval was obtained from the Saint Joseph hospital to access the data. All analyzed and published data were used anonymously without any identifying information.

Consent

The Ethics Committee of Saint Joseph Hospital waived the need for ethics approval and the need to obtain consent for the collection, analysis and publication of the retrospectively obtained and anonymized data for this non-interventional study.

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The study did not receive any financial help.

Author contribution

Data curation: M.D., S.Q., Y.A.A., E.A.A., Z.Z. Writing original draft: M.D., S.Q., Y.A.A., M.I., E.A.A., Z.Z. Conceptualization: Y.A.A., M.I., E.A.A., Z.Z.. Methodology: Y.A.A., E.A.A., Z.Z..

Formal analysis: E.A.A., Z.Z.. Writing—reviewing and editing: Y.A.A., M.I., E.A.A., Z.Z.

Conflicts of interest disclosure

The authors declare no conflicts of interest.

Research registration unique identifying number (UIN)

Not applicable.

Guarantor

Yousef Abu Asbeh.

Data availability statement

Dataset is available upon reasonable request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

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