### **ORIGINAL ARTICLE**



# Post-intubation tracheal stenosis in pediatric age group: single-center experiences of 24 years

 $Mojtaba\ Mokhber\ Dezfuli^1\cdot Seyed\ Reza\ Saghebi^2\cdot Mohammad\ Behgam\ Shadmehr^2\cdot Azizollah\ Abbasidezfouli^1$ 

Received: 9 November 2021 / Accepted: 21 December 2021 / Published online: 8 January 2022 © The Author(s), under exclusive licence to The Japanese Association for Thoracic Surgery 2022

# Abstract

**Background and aim** Post-intubation tracheal stenosis (PITS) is an iatrogenic injury that involves some patients. Given the importance of this issue and the referral of a significant number of children with tracheal stenosis to Masih Daneshvari Hospital in Tehran, Iran, the present study investigated tracheal stenosis following prolonged intubation in the pediatric age group. **Methods** In this observational retrospective study, from 1994 to 2018, the medical records of all children under 14 years of age with a history of PITS were reviewed. Demographic and clinical characteristics including signs and symptoms, the underlying condition that leads to intubation, duration of intubation, type of stenosis, and the therapeutic approach, type of surgery, and follow-up were collected and analyzed using SPSS.

**Results** Among 161 patients with a mean age of  $9.8 \pm 4.2$  years, 69% were male. The site of stenosis was limited to the trachea in 47% and others both trachea and subglottic area were involved. The most common cause of intubation was trauma. The most common symptoms were dyspnea and wheezing. Success rates of reconstruction were 93.75% in type I, 82.15% in type II, and 35.70% in type III. Among the 16 patients who underwent Type III surgery, decannulation was not performed in 11 patients. Traction in the anastomosis and complications were stated in 26 and 10% of the patients respectively, a mortality rate of 8.7% was also reported.

**Conclusion** In the case of endotracheal intubation, PITS should be considered in the differential diagnosis of dyspnea in children as well as adults.

Keywords Post-intubation tracheal stenosis · Pediatrics · Resection and anastomosis · Surgery

# Introduction

Airway management is the most important issue with endotracheal intubation, which is required in some patients with acute respiratory failure or trauma; however, this procedure may cause some complications. In this regard, tracheal stenosis secondary to intubation is a common acquired airway disorder [1, 2]. Other causes of acquired tracheal

<sup>2</sup> Tracheal Diseases Research Center (TDRC), National Research Institute of Tuberculosis and Lung Diseases (NRITLD), Shahid Beheshti University of Medical Sciences, Tehran, Iran stenosis include tracheostomy, direct trauma, tumor, and burn [3].

Mostly, pure tracheal involvement is less common in children, and most of these stenoses involve the subglottic area [4, 5]. PITS occurrence have been reported to a large extent, varies between 6 and 22% [6] and 3 and 8% [7]. In this regard, endotracheal tube size, the duration of intubation, traumatic intubation, the number of procedure, the route of intubation, the composition of the tube and cuff, and inflammation related to gastroesophageal reflux, bronchiolitis and viral infection are considered for airway stenosis [8].

In a prospective cohort study conducted by Schweiger et al., stridor was detected in 44.38% of the pediatric patients with the 59.18% specificity according to laryngoscopy. In case of mild and transient post-extubation stridor, bronchoscopy is not mandatory. But, sever, late onset or progressive stridor require more investigation [9]. Our 24-year experiences revealed that 7% of all registered PITS are occurred in pediatric age group.

Seyed Reza Saghebi sr\_saghebi@yahoo.com

<sup>&</sup>lt;sup>1</sup> Lung Transplantation Research Center (LTRC), National Research Institute of Tuberculosis and Lung Diseases (NRITLD), Shahid Beheshti University of Medical Sciences, Tehran, Iran

Tracheal injury has a wide spectrum from mucosal damage and inflammation to granulation formation, destructive changes in the cartilage tissue, and finally tracheal narrowing. The exact cause of PITS is unknown, although some factors including duration of intubation, cuff pressure in case of using cuffed endotracheal tubes, and local infection have been discussed [6].

The patient's signs and symptoms are related to the tracheal lumen narrowing and include a wide range from asymptomatic condition to severe stridor, dyspnea, and death [10]. Almost, when the diameter of the tracheal lumen reaches 50% the patient becomes symptomatic [11]. And, it is considered that in more than 40% of all cases, the first diagnose of symptomatic patients is asthma [10, 12]. Mild stenosis in patients may result in recurrent pneumonia and progressive dyspnea [13]. Hence, in all children with respiratory symptoms and a history of endotracheal intubation, the possibility of stenosis should be considered.

From a therapeutic approaches point of view, tracheal resection-anastomosis is recommended based on feasibility [13]. However, when we face a long segment of stenosis i.e. one-third of the total tracheal length, the anastomosis site will be prone to dehiscence, Therefore, other treatment options such as dilatation via rigid bronchoscopy and laser therapy or stenting should be considered. Each of these options has its limitations and sometimes tracheal reconstruction using an autograft or allograft is the last plan [14]. In the report from Yamamoto et al. of 45 children out of 141(31.9%) required laryngotracheal reconstruction [15] and 24% restenosis was observed with mortality rate of 4.4%.

With respect to the high prevalence of tracheal stenosis in our society according to the **Alborz database** (more than 2000 patients in 20 years of data registry) [6] and specific consideration in children, in the present study, we aimed to evaluate tracheal stenosis following prolonged intubation and the results of surgery in the pediatrics. We focused experience in the management of this very much challenging and uncommon complication that could be useful in the field of tracheal surgery.

We decided to report our updated,

# **Materials and methods**

## **Study design**

This retrospective observational study was performed on all patients with endotracheal stenosis after intubation in the pediatric age group who were treated at Masih Daneshvari Hospital, from 1994 to 2018. The files that were registered in the Research Institute of Tuberculosis and Lung Diseases of Masih Daneshvari Hospital were included in the study after in case of meeting the inclusion criteria.

#### Inclusion and exclusion criteria

All patients registered at the Alborz database (under 14 years of age) with tracheal stenosis due to intubation.

The primary object was decannulation of the airway with no tracheostomy or T-tube.

# **Data collection**

The medical record of the patients with tracheal stenosis following prolonged intubation were reviewed; then, the demographic and clinical information including clinical symptoms, cause of intubation, duration of intubation, complications, treatment outcome and mortality, and other required information were extracted and entered in the study information form.

#### **Treatment approach**

Types of interventions performed included resection and anastomosis of the trachea (type I), resection and anastomosis of the trachea, and part of the anterior cricoid arch (type II), and resection and anastomosis of the trachea part of the cricoid and posterior and/or anterior laryngofissure (type III). In some patients, no resection treatment was performed due to the improper condition of the larynx and they underwent supportive treatments such as tracheostomy or T-tube implantation.

#### **Data analysis**

After the data collection, study groups were compared employing SPSS software version 16. Descriptive variables were presented as frequency and percentage, quantitative variables were presented as mean  $\pm$  SD. Based on the types of comparison, chi-squared and *t* test and oneway ANOVA were performed considering a significance level of 0.05.

# **Ethical considerations**

This study was approved by the Ethics Committee of the university. Confidentiality of the patient's name and details and observance of Helsinki provisions, as well as observance of ethical principles in collecting library information, were considered in all stages of the study.

## Results

Among the 161 children who had tracheal stenosis following prolonged intubation, 69% were boys (age:  $9.8 \pm 3.6$  years) and 31% were girls (age:  $8.4 \pm 4.2$ ), with no considerable difference in the age in different genders, (P = 0.054).

63% of children were under 12 years old; the mean age of cases was  $9.8 \pm 4.2$  years with the range of 4 months to 14 years (Table 1).

Regarding symptoms: the most complaint was dyspnea with the frequency of 53% and at the second place, wheezing was considered in 20% of the patients (Table 2).

 Table 1 Demographic information of the children with tracheal stenosis following prolonged intubation

Demographic characteristics	Frequency	Percentage	P value	
Gender				
Female	49	31%	0.25	
Male	112	69%		
Age				
Under 1 year	3	1.8%	0.95	
1–2 years	11	6.8%		
3–5 years	26	16%		
6–8 years	27	16.7%		
9–11 years	35	21.7%		
12–14 years	59	36.6%		
Cause of intubation				
Accident	105	65.2%	0.18	
Trauma and injuries to the head	10	6.2%		
Falls from a height	12	7.4%		
Burn	4	2.5%		
Suicide	3	1.8%		
Others	13	8%		
Not mentioned	14	8.6%		
Diagnosis				
Stenosis following intubation	157	98.5%	0.45	
Airway injury	4	1.5%		
Site of involvement				
Trachea	76	47.2%	0.002	
Trachea and subglottis	85	53.8%		
Duration of intubation				
1–14 days	59	36.5%	0.47	
15–20 days	36	21.5%		
21–25 days	8	4%		
26–30 days	11	9%		
More than 30 days	47	30%		
Type of intervention				
Type I	32	42%	0.04	
Type II	28	38%		
Type III	16	20%		

Symptoms	Frequency	Percentage
Shortness of breath	84	53
Wheezing	32	20
Fever and seizures	7	5
Decreased level of consciousness	4	3
Severe respiratory distress and restlessness	3	2
Others	27	17

# **Intubation etiology**

The most common causes of intubation were trauma and injuries to the head (65.2%), falls from a height (13.6%), burns (2.5%), and suicide (1.8%), other causes were Guillain–Barre syndrome, history of asthma, and leukemia, and each of them was observed in 2 cases, with no significant difference, (P=0.18).

#### Involved area

In 76 patients (47%), the site of involvement was only the trachea, and in 85 patients (53%) the site of involvement was the trachea and subglottis are. Involvement of subglottic are accompanied with trachea was significantly more than pure trachea involvement (P = 0.002). There was no significant difference regarding age, gender and days of intubation in pure tracheal involvement and subglottic stenosis (Table 3).

## **Duration of intubation**

In most patients (36%) the duration of intubation was 1-14 days. Also, there was a history of more than 30 days intubation period in 30% of the patients. There was no significant difference between groups of intubation-duration (P=0.47).

Regarding type of surgery, there was no significant differences in duration of intubation. In type I, II, III and IV, intubation time was  $17.4 \pm 11.2$ ,  $14.10 \pm 7.9$ ,  $15.8 \pm 10.1$  and  $11.6 \pm 5.7$  days respectively, (F = 0.47, P = 0.7).

#### The location of stenosis and types of intervention

In total, 71 patients had a tracheostomy before getting treatment, and 5 had a T-tube. Type I procedure was performed in 42% of patients, and type II was used in 38% of patients. Among the studied variables, only the site of involvement and the type of intervention had a significant relationship with tracheal stenosis (P < 0.05).

According to Fig. 1, which shows the frequency of interventions performed in all patients with pure tracheal

Table 3Involved area andthe type of surgery in theairway among PITS patients inpediatric aged group



Fig. 1 Type of intervention in different locations of stenosis

stenosis, 38 patients (29: type I and 9: type II) underwent surgery, 13 patients underwent T-tube implantation and 13 patients underwent tracheostomy. 28 patients were discharged in good general condition without stenosis and hoarseness.

According to the frequency of surgeries performed in patients in which the site of involvement was trachea and subglottis, 38 patients (3: type I, 19: type II, and 16: type III) underwent surgery, 21 patients underwent tracheostomy, and 4 patients underwent T-tube implantation. Patients who underwent type III surgery were usually discharged with a T-tube or tracheostomy stand.

#### **Recurrences of stenosis**

A total of 10 re-stenosis were reported, of which 8 patients were treated with dilatation, and corticosteroids, and tracheostomy were performed for two patients. No recurrence of stenosis was reported in patients who underwent type I surgery. Out of 20 patients who underwent type II surgery, 3 patients had a recurrence of stenosis, and during follow-up, one patient had to undergo tracheostomy.

General Thoracic and Cardiovascular Surgery (2022) 70:553–558

## **Decannulation and complications**

Among the 16 patients who underwent type III surgery, decannulation was not performed in 11 patients. The success rate in type I operations was 93.75%, in type II was 82.15%, and in type III operations was equal to 35.70% (Table 4).

Based on the operation description, it was shown that 42 patients (26%) of the total patients had traction in the anastomosis. 10% of patients had complications including vocal cord injury, vocal cord paralysis, hemorrhage, site seroma, site hematoma, and need for repeated dilatation. A total of 14 deaths were reported, of which 8 were related to suffocation due to surgery (suffocation, injury, and tracheal rupture), and the exact causes of death in the remaining cases were not known based on the medical records.

The duration of intubation in survived and not survived cases was  $15.8 \pm 101$  and  $15.22 \pm 9.2$  days (P = 0.87). Also, there was no significant differences between in survived and not survived groups. Indeed, mean age was  $9.60 \pm 3.9$  and  $9.19 \pm 3.3$  years in these patients respectively, (0.773).

# Discussion

Despite efforts to prevent and reduce the complications of endotracheal intubation, tracheal intubation and its complications still cause many respiratory problems for patients [6]. Our study inconsistent with the other studies

Table 4The rate ofdecannulation after treatment oftracheal stenosis according tothe type of surgery in pediatricage group

Type of surgery	Number of patients	Airway decannulation N (%)	Not decannulation N (%)	P value
Туре І	32	30 (93.75)	2 (6.25)	0.07
Type II	28	23 (82.15)	5 (17.85)	
Type III	16	5 (35.70)	11 (64.3)	

showed that the most common cause of prolonged intubation was vehicle accident and then brain injury [16].

In our study, the most common site of involvement was the subglottis with the trachea. Similar studies also showed that in children, the most common site of involvement was the subglottis in addition to the upper trachea [17]. A study by Meneghini et al. on the risk factors for subglottic stenosis following prolonged intubation in children showed that the incidence of subglottic and tracheal stenosis in children was higher than in adults [18]. In addition, the duration of intubation is an important factor in endotracheal stenosis, although, in our study, 36% of the patients reported one to 14 days of intubation, on the other hand, intubationduration was more than 30 days in 30% of people.

Our study revealed that decannulation is poor in the case of type III surgery rather than type I and type II. In this regard, a study by Abbasi et al. showed that resection procedure had excellent efficacy in 52 patients (74.2%), good efficacy in 9 patients (12.8%), and acceptable efficacy in 6 patients (8.5%), but in 3 cases the surgery resulted in death (4.2%) [19]. Marstone et al. showed that resection had appropriate results for grade III and IV subglottic stenosis. This method has no harmful effects on the growth and function of the larynx, and sound quality improves significantly after surgery, however, largely depends on preoperative conditions. Other studies have also reported that surgical treatment is effective in reducing tracheal complications [20, 21]. Monnier et al. examined 38 infants and children with severe subglottic stenosis undergoing surgery. No laryngeal nerve damage and no mortality were observed among the cases. The 10-year follow-up showed that all patients had normal growth of the larynx and trachea. The results of this study showed that, in comparison with laryngotracheoplasty, the resection method has good efficacy for the treatment of subglottic stenosis. They stated that this operation should be selected for patients with severe stenosis (grade III and IV) [22].

Providing endotracheal intubation training programs can positively affect the performance of health professionals. A recent study in this regard considered poor skill levels as one of the causes of tracheal stenosis after intubation and stated that training is needed to prevent complications and reduce cases of tracheal stenosis [23].

In general, early consultation with surgeons who have great experience in the treatment of cases with stenosis is recommended for better treatment planning.

To minimize the complications regarding laryngotracheal resection and reconstruction, two key points including tension and devascularization should be managed. Resections greater than 4 cm should be considered to reduce anastomotic tension. Some techniques such as pretracheal plane dissection, neck flexion to avoid head extension for 5 days, traction sutures and suprahyoid laryngeal release in case of long stenosis is recommended [21].

In addition, careful planning and preparation of a stenosis prevention chart can be useful in the intensive care unit. Also, airway management workshops and classes for nurses and caregivers of intubated patients in the intensive care unit can be effective in reducing the incidence of this complication.

One of the limitations of this study was the incomplete file of some patients, which is one of the disadvantages of retrospective studies, and it was eliminated as much as possible through telephone coordination and information review. Also, only demographic characteristics and duration of intubation was considered for location of the stenosis and other factors including endotracheal tube characters, infection during ICU admission, and the care of the tube and so on were not assessed as the background for the level of involvement.

# Conclusion

In the case of endotracheal intubation, PITS should be considered in the differential diagnosis of dyspnea in children as well as adults. The frequency of subglottic involvement in tracheal stenoses after intubation is high in children, but similar to adults, if there is no subglottic involvement, most patients are treated completely by resection and anastomosis. In cases of subglottic and laryngeal involvement, decannulation rate is lower than pure tracheal involvement and complex surgical treatments are required, such as the reconstruction of the cricoid cartilage and larynx, which in many cases do not induce the desired results. Certainly, acquaintance with the strengths and weaknesses of treatment strategies and further understanding of the effectiveness of new methods will be effective in preventing post-intubation tracheal stenosis and reducing related complications.

Author contributions All authors attest that they meet the current ICMJE criteria for authorship.

**Funding** This study was supported by National Research Institute of Tuberculosis and Lung Diseases.

#### Declarations

Conflict of interest The authors have no financial disclosures.

Ethical consideration IR.SBMU.NRITLD.REC.1399.070

### References

 da Costa Gaspar MT, Maximiano LF, Minamoto H, Otoch JP. Tracheal stenosis due to endotracheal tube cuff hyperinflation: a preventable complication. Autops Case Rep 2019;9(1):e2018072.

- Mehdizadeh J, Safikhani R, Langroudi MM. Laryngotracheal injury following prolonged endotracheal intubation. Tehran Univ Med J TUMS Publ. 2006;64(5):111–9.
- 3. Schweiger C, Manica D. Ongoing laryngeal stenosis: conservative management and alternatives to tracheostomy. Front Pediatr. 2020;8:161.
- 4. Graham JM, Scadding GK, Bull PD. Pediatric ENT. Berlin: Springer Science and Business Media; 2007.
- 5. Bibas BJ, et al. Predictors for postoperative complications after tracheal resection. Ann Thorac Surg. 2014;98(1):277–82.
- 6. Farzanegan R, et al. An overview of tracheal stenosis research trends and hot topics. Arch Iran Med. 2017;20(9):598–607.
- Li C, Rutter MJ. Acquired tracheal stenosis: cervical slide tracheoplasty. Semin Pediatr Surg. 2021;30(3): 151058. https://doi.org/ 10.1016/j.sempedsurg.2021.151058.
- Rutter M, Kuo IC. Predicting and managing the development of subglottic stenosis following intubation in children. J Pediatr (Rio J). 2020;96(1):1–3. https://doi.org/10.1016/j.jped.2019.04.001.
- Schweiger C, et al. Accuracy of stridor-based diagnosis of postintubation subglottic stenosis in pediatric patients. J Pediatr (Rio J). 2020;96:39–45.
- Spittle N, McCluskey A. Tracheal stenosis after intubation. BMJ. 2000;321(7267):1000–2.
- Wain JC. Postintubation tracheal stenosis. Chest Surg Clin. 2003;13(2):231–46.
- Sarkar NK, Kibria AA. Tracheal stenosis presenting as asthma following short-term intubation. J Bangladesh Coll Physicians Surg. 2021;39(3):205–8.
- Farzanegan R, Zangi M, Abbasidezfouli A, Pejhan S, Sadeghbeigee F, Daneshvarkakhki A, Sheikhy K, Saghebi SR, Nazemy S, Jahanshahi N, Shadmehr MB. Postintubation multisegmental tracheal stenosis: a 24-year experience. Ann Thorac Surg. 2021;112(4):1101–8.
- 14. Ghorbani F, Moradi L, Shadmehr MB, Bonakdar S, Droodinia A, Safshekan F. In-vivo characterization of a 3D hybrid scaffold

based on PCL/decellularized aorta for tracheal tissue engineering. Mater Sci Eng C. 2017. https://doi.org/10.1016/j.msec.2017.04. 150.

- 15. Yamamoto K, Monnier P, Holtz F, Jaquet Y. Laryngotracheal reconstruction for pediatric glotto-subglottic stenosis. Int J Pediatr Otorhinolaryngol. 2014;78(9):1476–9.
- Lee J-C, et al. Subglottic stenosis in children: our experience at a pediatric tertiary center for 8 years in South Korea. Int J Pediatr Otorhinolaryngol. 2019;121:64–7.
- Cuestas G, Rodríguez V, Doormann F, Munzón PB, Munzón GB. Endoscopic treatment of acquired subglottic stenosis in children: predictors of success. Arch Argent Pediatr. 2018;116(6):418–25.
- Acosta L, Cruz PV, Zagalo C, Santiago N. Iatrogenic tracheal stenosis following endotracheal intubation: a study of 20 clinical cases. Acta Otorrinolaringol Esp. 2003;54(3):202–10.
- Abbasidezfouli A, et al. Postintubation multisegmental tracheal stenosis: treatment and results. Ann Thorac Surg. 2007;84(1):211–4.
- Marston AP, White DR. Subglottic stenosis. Clin Perinatol. 2018;45(4):787–804.
- Axtell AL, Mathisen DJ. Idiopathic subglottic stenosis: techniques and results. Ann Cardiothorac Surg. 2018;7(2):299.
- 22. Monnier P, Lang F, Savary M. Cricotracheal resection for adult and pediatric subglottic stenoses: similarities and differences. Oper Tech Otolaryngol Neck Surg. 1999;10(4):311–5.
- Jefferson ND, Cohen AP, Rutter MJ. Subglottic stenosis. Semin Pediatr Surg. 2016;25(3):138–43.

**Publisher's Note** Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.