

**RESEARCH ARTICLE** 

# The Assessment of Congenital Laryngeal Lesions in Infants with Stridor

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#### ABSTRACT

Objective: In neonatal stridor, various conditions can be responsible as well as laryngomalacia. These conditions can be quite rare and the treatment of these diseases could be complicated. The purpose of this study is to identify the laryngeal pathologies and to discuss our approach in infants with chronic stridor.

Methods: The hospital charts of infants with stridor undergoing rigid laryngotracheobronchoscopy in our hospital in 2018-2022 were retrospectively reviewed.

**Results:** 107 children were enrolled to the study. The most frequent diagnosis was laryngomalacia (isolated and seen with secondary airway lesions (SALs)) (74 patients, 69.1%). 10 patients (9.3 %) had subglottic stenosis which caused stridor or dyspnea. Regarding other laryngeal lesions, in 8 patients (7.47 %) the diagnosis was laryngeal edema and 8 patients (7.47 %) had tracheomalacia. SALs which occurred with laryngomalacia were seen in 16 patients of our series (14.9 % of all cases). Surgery was performed in 11 of patients. Stridor was resolved in % 80 of laryngomalacia patients at about 2 years of age with only follow up.

**Conclusions:** In neonatal stridor, various conditions can be responsible as well as laryngomalacia. Referral of infant to otorhinolaryngologists and examination with flexible and rigid endoscopy is necessary for the assessment of stridor. The examination of the airway with the rigid endoscopy under general anesthesia without intubation on operating room with the cooperation of the anesthesiologist may provide the surgical intervention together with simultaneous inspection especially in the patients with severe stridor and systemic diseases.

Keywords: Stridor, Infant, Laryngomalacia, Stenosis, Endoscopy

#### INTRODUCTION

Stridor in pediatric patients is common symptom caused by turbulent air flowing through a narrowed airway (1,2). It can be inspiratory with obstruction at the supraglottic and glottic level, biphasic at the subglottic level and expiratory with obstruction in the trachea (1). Stridor in pediatric age group caused by viral croup, epiglottitis and foreign bodies represents acute stridor and requires emergent management. Chronic stridor is seen especially in neonatal and infantile period and usually caused by congenital laryngeal anomalies such as laryngomalacia, subglottic stenosis, vocal cord paralysis and subglottic hemangioma (2).

Laryngomalacia is the most seen congenital problem. During spontaneous respiration especially with inspiration, the supraglottic structures, specifically the epiglottis and aryepiglottic folds, close onto over the airway like a lotus flower due to their weak cartilage structure (3). The lesions that seen together laryngomalacia such as subglottic stenosis, tracheomalacia, vocal cord paralysis are called secondary airway lesions (SALs) (3,4). These lesions have clinical importance in several aspects. Firstly, surgical intervention may be required in cases of subglottic stenosis and cord paralysis that mostly seen ones. Secondly, even if surgical intervention is performed, it would be more appropriate to follow up for a period of time for accompanying laryngomalacia because of the nature of its. In this case, since stridor will continue even if it decreases, it is important to accurately diagnose whether only these lesions are present or whether they are present together with laryngomalacia in order to accurately predict the clinical outcome of the patient. Therefore, it is of great importance to perform an endoscopic examination of every patient who has stridor even if we guess likely having laryngomalacia to diagnosis SALs seen together with laryngomalacia. However, when conditions known to be associated with large airway lesions are taken into account, the prevalence of SALs

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associated with laryngomalacia is contrary to expectations. In fact, the prevalence of SALs is not fully revealed in the literature. The increased prevalence in some publications may be a result of the inclusion of patients with other pathologies and over-diagnosis for these lesions (5).

Most of congenital airway lesions such as laryngomalacia and tracheomalacia usually resolve by only follow up (2,6). However, rare congenital anomalies of airway such as subglottic stenosis, laryngeal webs, cysts and subglottic hemangioma require medical or surgical treatment (1,7). Presence of congenital laryngeal lesions and synchronous airway problems and various comorbidities are seen frequently in neonatal intensive care unit (2).

Management of a newborn with stridor, with or without respiratory distress, could be established with safe airway and evaluation of laryngeal structures (6). Flexible or rigid laryngoscopy can be used in otolaryngology departments easily for assessment of infant airway (6,8). Rigid laryngotracheobroncoscopy may provide the chance to intervene the pathology simultaneously during the examination. During rigid laryngotracheobroncoscopy under general anesthesia, if surgical intervention is required, the collaboration between the otolaryngologist and anesthesiologist is indispensable to ensure successful operative outcomes (9).

In this study, we have reviewed our experience in patients with chronic stridor requiring hospitalization and rigid endoscopy under general anesthesia. Our aim is to investigate whether management of pediatric patient with stridor with rigid endoscopy is always necessary.

#### MATERIAL AND METHODS

#### Patients

Ethical approval and Funding: This study was approved by Baskent University Institutional Review Board (Project no: KA19 / 412) and supported by Baskent University Research Fund.

The records of patients between 0-24 months of age who underwent laryngotrachobronchoscopy for chronic stridor between January 2018 and January 2022 in our tertiary hospital reviewed retrospectively. Cases with acute stridor caused by acute inflammation such as epiglottitis, croup, foreign bodies, trauma and oronasopharyngeal lesions such as choanal atresia were excluded.

### **Surgical Procedure**

Direct rigid laryngoscopy with/without tracheobronchoscopy was performed under general anesthesia in operating room. The patient was induced with either intravenous sedation with typically propofol (Propofol %1; Fresenius, Avusturya Gmbh., Avustria) or inhalational anesthetic with usually sevoflourane (Sevorane %100; Aesica, Queenborough Ltd., England) and then maintained at a proper depth anesthesia with spontaneous ventilation. Intubation was not performed. The laryngoscope was performed and 0° Storz-Hopkins telescope (ranging from

1.7 mm to 4 mm) was used for airway examination. The hypopharynx, larynx, subglottis, trachea and bronchi were evaluated systematically and documented photographically. When saturation drops below 80% during the procedure, the anesthetist warns the otorhinolaryngologist. In this case, the otorhinolaryngologist removes the endoscope and gives the patient to the anesthesiologist for ventilation and recovery of saturation. Then the procedure continues again. In this way, the airway is examined in detail.

The airway was sized with uncuffed endotracheal tube to determine the grade of stenosis according to Myer-Cotton classification (10). If surgical intervention was necessary, intubation was done and surgery was performed.

#### **Data Collection**

In each case, the following parameters were assessed: age at onset, sex, symptoms, diagnosis, synchronous airway lesions, presence of associated neurological and/or congenital anomalies, need for surgery and treatment modalities, negative situation during anesthesia, time of follow up, complications and time to resolution of symptoms.

The outcome of the patients in this study was classified as good outcome, poor outcome and death. The good outcome means normal daily life without any symptoms; the poor outcome means unresolved problems with/without tracheotomy.

#### **Statistical Analysis**

dvspnea

cyanosis

Statistical analysis (SPSS) (Version 22, Chicago IL, USA) was used for statistical analysis of the data. Chi-square test and Anova test was used for comparison of the two groups. Significance level in statistical evaluations was accepted as p<0.05.

#### RESULTS

A hundred and seven children were included in this study. There were 65 (60.7 %) male and 42 (39.2%) female children. The age of onset of symptoms was between 0-17 months and was within 12 months in 82 patients (76.6%). The most frequent symptom was stridor (58 patients, 54.2%) followed by dyspnea with 45 patients (42%) (Figure 1).

### symptoms

hoarseness feeding difficulties failure to thrive



Figure 1: Presenting symptoms in patients evaluated with stridor

The most frequent diagnosis was laryngomalacia (isolated and seen with secondary airway lesions) (74 patients, 69.1%). Ten of all patients (9.3 %) had subglottic stenosis causing stridor or dyspnea. Regarding other laryngeal lesions, there was laryngeal edema in 8 patients (7.47 %), tracheomalacia in 8 patients (7.47 %).

Figure 2 summarized the patients with diagnosis of laryngomalacia. Secondary airway lesions (SALs) which occurred with laryngomalacia were seen in 16 patients of our series (14.9 % of all cases). The most seen SAL was subglottic stenosis in 5 patients. Seventy-four patients had laryngomalacia and 60 of them was mild whereas 12 of them was moderate and 2 of them had more serious disease (Figure 3). Among the patients with laryngomalacia, 7 of them (7/74 9,4 %), were operated because of SALs and outcome of these patients was good. In 59 patients of this group (59/74, 80%) stridor resolved with only follow-up at the end of 36 months. Eight patients of them (8/74, 10,6%) had associated comorbidities with poor outcome.

Subglottic stenosis was seen in 10 patients (9.3%) and 5 of them have seen together with laryngomalacia and one of them had been undergone endoscopic laser operation with good outcome (Figure 4). Vocal cord paralysis was seen in 6 patients (5.6%) and 2 of them was managed with tracheotomy.

The presence of associated anomalies and/or diseases of the patients in this study were also recorded and the results were



# laryngomalacia with SALs

#### Figure 2: Diagrams of patients with Laryngomalacia



Figure 3: Severe isolated Laryngomalacia



Figure 4: Subglottic stenosis

given in Table 1. Twenty-eight patients (26.1%) had various comorbidities. The percentage of prematurity in this study was 39.2% (42 patients). Features of our patients were summarized in Table 1.

Our patients were followed- up during 2 years (2- 24 months). The surgery was performed in 11 patients (10.2%) in all series. Endoscopic surgery was performed in 9 of these patients (81,8%) with good outcome.

The outcome of the patients in this study was analyzed and shown in Table 2. Tracheotomy was applied in 7 patients (6.54%) and there were 9 deceased patients (8.41%) in our series. The infectious disease or comorbidity was the most common cause of death rather than laryngeal disorders. Tracheotomy was requiring mostly in the patients with comorbidities and the patients with tracheotomy had mostly poor outcome or deceased.

#### DISCUSSION

Laryngomalacia is the most common cause of newborn stridor and airway obstruction with an incidence of 35-75% (6). The most important symptom of laryngomalacia is inspiratory stridor which worsens at 4-6 months, improve at 8-12 months and will resolve by 12-18 months of age (6,10). The severity of symptoms of laryngomalacia are mild, moderate and severe. Severe form sometimes may require surgical management (6). In our study, the frequency of laryngomalacia was found as 69.1% and is compatible with literature. However, in our study there was no surgical treatment for laryngomalacia. Most of our cases were resolved by only follow-up at mean age of 17 months.

We separately evaluated secondary airway lesions seen together with laryngomalacia in this study. Because, even if we diagnose and treat these secondary lesions, unless the accompanying laryngomalacia improves, the patient's complaint decreases but will not go away, and this will affect the patient's outcome. Additionally, our goal is also to determine how often isolated laryngomalacia is seen. In fact, laryngomalacia is a condition improved by just waiting and growing of the patient. Thus,

N=107	Isolated Laryngomalacia	Laryngomalacia with SALs	Pathologies other than laryngomalacia	р
	(n= 58, 54,2%)	(n=16, 14,9%)	(n=27, 30,9%)	
Operating age (mean, mo)	11.2 months	9.7 months	12 months	>0.05
Gender (M/F)	45/13	10/6	14/13	>0.05
M (n=65, 60.7 %)				
F (n=42, 39.2%)				
Diagnosis				
<ul> <li>Subglottic stenosis</li> </ul>	-	5	5	
Vascular ring	-	2	-	
Tracheomalacia	-	2	6	
<ul> <li>Laryngeal web</li> </ul>	-	2	2	
Hemangioma	-	1	2	
<ul> <li>Cord paralysis</li> </ul>	-	2	4	
<ul> <li>Vallecular cyst</li> </ul>	-	2	-	
<ul> <li>Papillomatosis</li> </ul>	-	-	3	
<ul> <li>Tracheal stenosis</li> </ul>	-	-	2	
Granulation	-	-	1	
• Edema	-	-	8	
Comorbidities				
• Prematurity (n=42, 39,2%)	30	5	7	>0.05
• Systemic diseases (n= 28, 26,1%)				
CNS	6	2	2	>0.05
CVS	6	2	2	
Multipl Anomalies	2	2	-	
Down Syndrome	2	-	-	
Operation (n=11)				
Open surgery	-	2	-	
Endoscopic excision	-	4	4	
<ul> <li>Endoscopic dilatation and laser</li> </ul>	-	1	-	

M: Male, F: female, CNS: Central Nervous System, CVS: Cardiovascular System, SALs: Secondary airway lesions.

#### Table 2: Clinical outcome results in patients evaluated in this study.

		Tracheotomy (+) (n: 7), (6,54 %)	Tracheotomy (-)	р
Good outcome	Comorbidities (+)	-	46	-
(n: 80), (74,7 %)	Comorbidities (-)	-	34	
Poor outcome	Comorbidities (+)	3	14	>0.05
(n: 18), (16,8%)	Comorbidities (-)	-	1	
Exitus	Comorbidities (+)	2	5	>0.05
(n: 9), (8,41 %)	Comorbidities (-)	2	-	

this study may be guiding in terms of correct information to the family, especially in the primary care setting, and correct manipulation of a newborn with stridor. In our study, we found the frequency of secondary airway lesions as 14.9 % and the most seen anomaly with laryngomalacia was subglottic stenosis which had major significance. Therefore, we thought that every patient with congenital chronic stridor should be undergone laryngotracheobronchoscopy.

Laryngomalacia may occur as isolated or in association with any other anomalies of airway or other systemic diseases with an incidence of 7.5-64 % in literature (1). Krashin and et al (11) claimed that secondary airway lesions seen together with laryngomalacia were found as a low rate of 7.5 % and

had minor significance. They advocated that routine search of synchronous lesions was unnecessary in children with laryngomalacia (11). However, in literature many studies are present about importance of secondary airway lesions. Sakakura et al (2) found that 47.3% of their patients had at least one synchronous lesion and 56.4% had various comorbidities. Rifai et al (5) found the prevalence of SALs as 7.7% and claimed that the prevalence is much less than previously believed. However, they maintain their practice of full airway examination for all patients with severe symptoms.

Fiberoptic flexible endoscopy has been become as preferred diagnostic tool in the congenital stridor nowadays (8). Dynamic anatomical motion and distortion of the airway's structures during breathing like laryngomalacia can be evaluated with fiberoptic endoscopy (11). However, rigid laryngoscopy may be superior in detailed evaluation of airway anatomy especially in examination of the posterior glottis area and subglottic stenosis (7,11). Rigid laryngotracheobroncoscopy is also necessary for diagnosis and management in many airway lesions that can be seen together with laryngomalacia (12). We thought that the preference of fiberoptic or rigid endoscopy in congenital stridor is dependent on the severity of the patient. If the infant has severe stridor with comorbidities such as cardiovascular disease, failure of thrive; the examination of the airway with the rigid endoscopy under general anesthesia without intubation in operating room may be safer than fiberoptic examination under policlinic conditions without anesthesia. However, of which diagnostic tool for laryngeal examination of infant with stridor is used, nasal and nasopharyngeal examination must be done due to significant airway compromise in neonates with nasal obstruction (12,13).

In many tertiary care pediatric hospitals, the patients with stridor are managed by a multidisciplinary team comprised of otolaryngologists, pediatrist, anesthesiologists, nutritionists, speech pathologists, and nurses. Management includes both medical optimization and surgical interventions, including endoscopic and open airway surgery. Preoperative optimization of the patient's comorbidities is paramount for favorable surgical outcomes (9).

Many comorbidities and many systemic diseases can be seen together with congenital airway diseases (2,7). Moreover, the infants with congenital airway diseases are susceptible to many infectious diseases which may be directly cause of death (2,12). In this study, 26 % of patients (28 patients) had suffered from systemic diseases and this condition affected the clinical outcome (Table 2). The most common comorbidity together with congenital airway diseases in our study was cardiovascular anomalies and central nervous system anomalies. Sakakura et al (2) also found that cardiovascular diseases were the most seen comorbidities in their patients with a percentage of 51 %. Prematurity can be also seen as comorbidity and is blamed for many airways congenital lesions (1,14). Martins et al (14) found that 27.3% of 55 patients were premature and Yuen et al (1) showed that 4 premature infants were present in their series of 26 children. However, Aksoy et al (15) said that prematurity and intubation were not absolutely essential for congenital airway cysts. Our prematurity rate was 39% and we thought also that congenital airway lesions were not absolutely together with prematurity.

The association of gastroesophageal reflux (GER) with congenital airway lesions especially laryngomalacia is well investigated in the literature (1,6). GER was found as responsible for many severe symptoms of laryngomalacia and other airway lesions (6, 16). Yuen et al (1) documented that the incidence of GER with laryngomalacia was 42.3%. In our series, unfortunately the frequency of GER in congenital airway lesions was not examined.

In our series 10.2 % of patients (11 patients) had been undergone operation because of congenital airway anomalies.

These operations were usually endoscopic surgeries and were performed for secondary airway lesions (Table 1). Any patient with isolated laryngomalacia had not been undergone surgery in our study. Because most of the isolated laryngomalacia patients in our study improved with time. Indeed, our patients were followed- up by 2 years and in 80 % of them, stridor resolved with only follow-up at the end of 24 months. In this series, patients with laryngomalacia or secondary airway lesions had also severe comorbidities and had usually tracheotomy. Tracheotomy and comorbidities were primary responsible from our patient's outcome (Table 2). 9 patients (8.41%) had deceased because of systemic primary diseases. Neither laryngomalacia nor secondary airway lesions seen together with laryngomalacia has directly affected the outcome in our study as similar in the literature.

In our study, we found that the most important cause of death was comorbidities or infection while investigating the most frequently seen congenital airway lesions. Sakakura et al (2) found that tracheotomy was one of the most related factors for outcome regardless of congenital laryngeal lesions. Nisa et al (17) concluded in their study that if the newborn with bilateral vocal cord paralysis had major comorbidities affecting their normal development, they had poor functional outcomes and would be tracheotomy-dependent. According to our results, we thought also, tracheotomy determined the end-results. However, in patients with tracheotomy, rules of tracheotomy surgery and the ability and knowledge of nurse in intensive care units were very important. These factors could be cause of elongation of period with tracheotomy and end result of the outcome. This opinion requires proof with future studies.

Vascular ring is aberrant right subclavian artery and also can be rare cause of congenital chronic neonatal stridor (17). In literature, it is pointed that congenital anomalies of aortic arch and its branches are rare but important causes of stridor and can be managed with surgery. In our series, 2 patients with vascular ring were operated with good outcome. Adamczuk et al (18) concluded that chronic congenital stridor was an interdisciplinary problem. In this regard, Asha'ari ZA et al (19) claimed that combined physician-surgeon airway endoscopy gave a high diagnostic yield and provided more efficient management with severe airway problems. In our hospital we did not apply endoscopy with pediatricians at the same time. However, all patients with chronic stridor were evaluated and consulted with pediatricians and were examined for circulatory and central nervous system anomalies as a cause of stridor.

The most important weak point of the study is that some information such as reflux and tracheotomy care could not be obtained due to a retrospective nature. Despite this weakness, this study offers a comparative analysis about congenital stridor.

#### CONCLUSION

As a conclusion, in neonatal stridor, various conditions can be responsible as well as laryngomalacia. Referral of all infants with stridor to otorhinolaringologists and the examination with flexible and rigid endoscopy is necessary for the assessment of airway. The examination of the airway with the rigid endoscopy under general anesthesia without intubation in operating room may provide simultaneous inspection and surgical intervention especially patients with severe stridor and with systemic diseases. It is thought that a multidisciplinary approach is much more important in stridor management rather than evaluating the airway with a rigid or flexible endoscope.

Ethics Committee Approval: This study was approved by the ethics committee of Baskent Univercity Institutional Review Board (Project no: KA19 / 412)

Informed Consent: Written consent was obtained from the participants.

Peer Review: Externally peer-reviewed.

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