original article

Outcomes of flexible fiberoptic laryngoscopy in patients with stridor: a cross-sectional study in a tertiary care pediatric center in Saudi Arabia

Ghada Alsowailmi,[a] Jaber Alshammari,[b] Abdullah Saud Arafat,[b] Amal Alotaibi,[a] Afnan Alsahli,[a] Sara Ibrahim Alshahwan,[a] Aamir Omair,[c] Maryam Alsafi[a]

From the [a]College of Medicine, King Saud bin Abdulaziz University for Health Sciences, Riyadh, Saudi Arabia; [b]Department of Pediatric Otorhinolaryngology, King Abdulaziz Medical City, Riyadh, Saudi Arabia; [c]Department of Medical Education, College of Medicine, King Saud bin Abdulaziz University for Health Sciences, Riyadh, Saudi Arabia

Correspondence: Dr. Ghada Alsowailmi · College of Medicine, King Saud bin Abdulaziz University for Health Sciences, Riyadh 11426, Saudi Arabia · ghada.alsowailmi@gmail.com · ORCID: https://orcid.org/0000-0001-5457-0576

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BACKGROUND: Successful evaluation of a patient with stridor requires a thorough history and physical examination followed by a flexible fiberoptic laryngoscopy (FFL), which provides visualization of the upper airway.

OBJECTIVES: Estimate the prevalence of causes of stridor in children who underwent FFL and compare different age groups. Find any significant associations between symptoms and laryngoscopic findings. Identify patients who needed further evaluation using direct laryngobronchoscopy (DLB).

DESIGN: Retrospective, cross-sectional.

SETTING: Tertiary care center in Riyadh.

PATIENTS AND METHODS: We included all pediatric patients aged 1 month to 14 years who underwent fiberoptic laryngoscopy for stridor evaluation from January 2015 to January 2018 (37 months). Patients older than the age of 14 years, and patients with a workable diagnosis with adenotonsillar hypertrophy, choanal atresia, or laryngotracheobronchitis (croup) were excluded.

MAIN OUTCOME MEASURES: Findings of FFL.

SAMPLE SIZE: 217 pediatric patients.

RESULTS: The median (interquartile range) age of the patients was 5 (8) months. Laryngomalacia was the most common diagnosis (n=149, 69%) followed by laryngopharyngeal reflux (n=42, 19%). Subglottic stenosis was the most common finding in patients who underwent DLB for further evaluation (n=19, 49%). Laryngomalacia was more frequent in children ≤12 months of age (83% vs 43% in children >12 months, *P*<.001). Vocal cord paralysis was more common in children >12 months of age (27% vs 9%, *P*<.001). FFL was effective in finding the diagnosis in 178 (82%) patients; only 39 (18%) patients needed further assessment using DLB.

CONCLUSION: FFL is an effective and important tool for evaluating patients with stridor.

LIMITATIONS: Retrospective design and single-centered.

CONFLICT OF INTEREST: None.

Stridor is a serious symptom that reflects airway obstruction and requires prompt investigation. Although there are numerous causes (inflammatory, traumatic, iatrogenic, congenital, and neoplastic origin), the most common cause of chronic stridor is laryngomalacia, while croup is the most common cause of acute stridor.[1] Successful evaluation of a patient with stridor requires a thorough history and physical examination. In addition, flexible fiberoptic laryngoscopy (FFL) provides visualization of the upper airway up to the vocal cord and requires only local anesthesia.[2] Silberman et al were the first to use the flexible fiberoptic scope under local anesthesia to investigate stridor in infants.[3] Since the flexible fiberoptic scope does not evaluate the trachea or subglottic area, which might be the primary site of obstruction, the gold standard for the diagnosis of upper airway problems is direct laryngobronchoscopy (DLB). However, DLB is associated with potential morbidity and mortality related to general anesthesia.[2]

FFL has been widely used to investigate the underlying causes of upper airway obstruction such as laryngomalacia, vocal cord palsy, epiglottitis, gastroesophageal reflux disease (GERD), and airway foreign bodies. A prospective study at Columbus Children's Hospital, Columbus, Ohio, found that of 64 children with stridor, 22 had subglottic stenosis, 9 had laryngomalacia, 8 had an airway foreign body, 6 had a normal airway, and 4 had tracheal stenosis.[2] FFL, DLB, and air-fluoroscopy were used to evaluate the patients. A study performed at Addenbrooke's Hospital, Cambridge, UK, included 66 infants who underwent per-oral FFL. The vast majority of patients were male (59%), under the age of 6 months, and their main complaint was stridor from birth (94%). Seven patients were unable to undergo FFL, and instead underwent microlaryngoscopy, while the remaining 59 patients underwent FLL. The most common finding was laryngomalacia (52%).[4] Another study, carried out between January 1998 and August 1999, included 43 patients who underwent FFL; in this study, laryngomalacia was the most common diagnosis (35 patients), vocal cord palsy was reported in 6, and a normal larynx was found in 2 infants.[5] After reviewing the local literature in Saudi Arabia, we found no descriptive statistics outlining the etiology of stridor diagnosed using FFL. Therefore, this study aimed to identify the causes of stridor in patients investigated by FFL at a tertiary care hospital in Saudi Arabia.

PATIENTS AND METHODS

The study was conducted in the King Abdullah Specialist Children's Hospital (KASCH), Riyadh, Saudi Arabia, which is considered one of the major tertiary referral centers. The center receives referrals from Saudi Arabia and all Arabian Gulf countries. The Otolaryngology Head and Neck Surgery department includes eight consultants and four associate consultants. In this retrospective cross-sectional study, we reviewed the charts of 4650 patients that presented with stridor and were evaluated using fiberoptic laryngoscopy. Investigators reviewed paper and electronic charts to extract the data. All pediatric patients (both sexes, all nationalities), ranging in age from 1 month to 14 years who underwent fiberoptic laryngoscopy for stridor evaluation from January 2015 to January 2018 and were followed up at KASCH were included. All patients older than the age of 14 years, and patients with a workable diagnosis with adenotonsillar hypertrophy, choanal atresia, or laryngotracheobronchitis (croup) were excluded. Ethical approval of the study was provided by the King Abdullah International Medical Research Center (KAIMRC) under protocol number RC19/113/R.

Variables included patient demographics, signs, symptoms, and past medical history. The outcome variables were the findings of flexible laryngoscopy. Data entry was managed using MS Excel, and the data were analyzed using IBM SPSS version 21. The results are presented as descriptive statistics with appropriate summary measures for continuous numerical variables, and as frequencies and percentages for categorical variables. The chi square test was used to compare the categorical variables. $P<.05$ was considered a statistically significant difference.

RESULTS

Of pediatric patients who underwent FFL over a period of 37 months (January 2015–January 2018), 217 presented with stridor. The median (interquartile range) age of patients was 5 (8) months (**Table 1**).

Twelve different findings were identified using a flexible laryngoscope (**Table 2**). Findings were observed in 203 (94%) patients, while 14 (6%) had no abnormalities. Of all diagnoses, laryngomalacia was the most commonly encountered (n=149, 69%) patients, followed by laryngopharyngeal reflux (n=42, 19%) (**Table 2**). In patients ≤12 months old (n=140), the difference in frequency of laryngomalacia compared with older children (n=33) was statistically significant ($P<.001$) (**Table 3**).

While vocal cord paralysis was more common in children >12 months of age (n=21, 27% of children >12 months) ($P<.001$), differences in the frequency of laryngopharyngeal reflux ($P=.078$) and supraglottic edema ($P=.349$) were not significantly different, even though they were among the most common findings (**Table 3**).

Vocal cord paralysis was significantly more likely to be present in girls (n=21, 21% of females) than boys (n=13, 11% of males) (*P*=.046) (**Table 4**). Moreover, patients diagnosed with vocal cord paralysis were more likely to have a past medical history of ICU admission (*P*=.051) and intubation (*P*<.001) (**Table 4**). This analysis was applied to all the other laryngoscopy findings, but none of the associations were found to be statistically significant. Weak voice and hoarseness were significantly more likely to be the presenting complaints of vocal cord paralysis (*P*<.001) (**Table 5**). Among all the symptoms assessed in the current study, positional changes, failure to thrive, and hoarseness were significantly associated with laryngomalacia (*P*<.001, .013, and .006, respectively). Laryngopharyngeal reflux was significantly more likely to be diagnosed in patients who presented with swallowing and feeding difficulties (*P*= .019) and in patients who presented with cyanosis (*P*= .017). Among the 217 patients included in this study, 39 (18%) patients required further evaluation using DLB to either diagnose or confirm their anticipated diagnosis. Subglottic stenosis was seen in the majority of these patients (n=19, 49% of patients who underwent DLB), while 13 (33%) patients had no other abnormal findings than those diagnosed with flexible laryngoscopy (**Table 6**).

Table 1. Characteristics of the subjects (n=217)

Age (months)	
12	140 (65)
>12	77 (35)
Sex	
Male	117 (54)
Female	100 (46)
Nationality	
Saudi	210 (97)
Non-Saudi	7 (3)
Presenting symptom/sign	
Stridor	217 (100)
Hoarseness	14 (7)
Weak voice/cry	30 (14)
Cough	18 (8)
Positional changes	60 (28)
Snoring	32 (15)
Sleep apnea	17 (8)
Cyanosis	24 (11)
Retraction	18 (8)
Tachycardia	13 (6)
Swallowing and feeding difficulties	75 (35)
Drooling	8 (4)
Failure to thrive	36 (17)
Current upper respiratory infection	39 (18)
Medical history	
ICU admission	107 (49)
Intubation	99 (46)
Respiratory diseases	38 (18)
Congenital anomalies	88 (41)
Cerebral palsy	24 (11)
Gastroesophageal reflux disease	76 (35)
Previous respiratory infection	104 (48)

Data are n (%).

DISCUSSION

This study provides the findings of fiberoptic laryngoscopy in the evaluation of stridor in children at a tertiary care hospital. We found that laryngomalacia and laryngopharyngeal reflux were the most prevalent diagnoses. Laryngomalacia was encountered more frequently in infants, while vocal cord paralysis was more common in patients >12 months of age. Patients with vocal cord paralysis were more likely to have a history of intubation and ICU admission. The presentations most often associated with laryngomalacia were positional change,

Table 2. Flexible laryngoscopic findings (n=217).

Laryngomalacia	149 (69)
Laryngopharyngeal reflux	42 (19)
Vocal cord paralysis	34 (16)
Supraglottic edema	15 (7)
Pharyngeal hypotonia	5 (2)
Nasopharyngeal mass	2 (1)
Asymmetrical arytenoid	2 (1)
Retropharyngeal abscess	1 (0.5)
Vallecular cyst	1 (0.5)
Foreign body	1 (0.5)
Laryngeal web	1 (0.5)
Nasal adhesion	1 (0.5)
No abnormalities	14 (6)

Data are n (%). Total more than 217 because more than one finding in some patients.

failure to thrive, and hoarseness. Vocal cord paralysis was associated with hoarseness and weak voice, while laryngopharyngeal reflux was associated with cyanosis and swallowing and feeding difficulties. The practice at our hospital is to use less invasive procedures in the evaluation of stridor; however, some patients required further evaluation with DLB, in which subglottic stenosis was the predominant finding.

In the present study, laryngomalacia was the most common diagnosis, which is similar to a previous study by O'Sullivan et al who found that 50% of stridor patients had laryngomalacia.[6] The second most common diagnosis in our study was laryngopharyngeal reflux, which is an extra-esophageal manifestation found in 60% of patients with GERD.[7] GERD is a well-established predisposing factor for airway compromise that causes inflammation and edema of the posterior glottic and supra-arytenoid tissue and is a well-known component of the pathological course of laryngomalacia.[8] In our study, 156 (72%) of laryngomalacia patients and 37 (17%) vocal cord paralysis patients had GERD. However, the *P* value was not statistically significant for these associations, probably due to the small sample size. One previous study by Erdem evaluated 109 patients with stridor for associated comorbidities and found that 31 patients had GERD.8 Another study by Kuo et al found that 7 out of 15 patients with vocal cord paralysis patients had gross reflux esophagitis and bronchoalveolar lavage positive for lipid laden macrophages.9 A study by Zoumalan et al found laryngomalacia in 73% of 202 patients with stridor aged <12 months,10 which is comparable to our finding that 83% of patients <12 months who were diagnosed with laryngomalacia. Another study by Nussbaum et al evaluated 297 laryngomalacia patients and classified laryngomalacia as type 1 if laryngeal collapse was the only scopic finding and type 2 if the patient had additional findings besides laryngeal collapse. They concluded that laryngomalacia type 1 was more common in early infancy, while laryngomalacia type 2 was associated with old age.[11] Our findings could not support their conclusions, as our data did not specify laryngomalacia by type. In our study, vocal cord paralysis was more common in patients >12 months, which is contrary to a study by Rosin that found the majority of vocal cord paralysis in patients aged <12 months.[12]

Zoumalan et al found that a primary diagnosis of laryngomalacia was more frequent in boys (in 202 patients, 101 patients with laryngomalacia were boys and 56 were girls).[10] In another study by Holinger et al that included 209 patients with stridor as their main complaint, 150 were male and 69 were female.[13] In the present study, the rate of laryngomalacia showed a slight male predominance, but the difference from females was not statistically significant. Our study revealed a significant sex difference in vocal cord paralysis: more patients were female (n=21, 21%) than male (n=13, 11%); the reason for this discrepancy remains equivocal. Similarly, a study by Nemer et al of vocal cord paralysis cases found that the majority of the patients were female (36 females compared to 17 males).[14]

A previous study by Kuo et al found that the most prominent presentations of congenital vocal cord paralysis were stridor and respiratory distress, with 26% requiring tracheostomy.[9] This is consistent with our study, as patients who were diagnosed with vocal cord paralysis were more likely to be intubated and admitted to the ICU compared to those with other causes for stridor[9]

In the present study, the most common symptoms, beside stridor (which was one of the inclusion criteria), were swallowing and feeding difficulties (n=75, 35%), positional changes (n=60, 28%), and failure to thrive (n=36, 17%). However, our findings were opposite those of another study that also had stridor as the main presenting symptom for all patients, but concluded that

Table 3. Association between selected laryngoscopic findings and patient age group.

Laryngoscopy findings	Age group (in months)		*P* value
	≤12 (n=140)	>12 (n=77)	
Laryngomalacia	116 (83)	33 (43)	<.001
Vocal cord paralysis	13 (9)	21 (27)	<.001
Laryngopharyngeal reflux	32 (23)	10 (13)	.078
Supraglottic edema	8 (6)	7 (9)	.349
No abnormalities	6 (4)	8 (10)	.08

Data are n (%). Statistical comparisons by chi-square test.

Table 4. Association between vocal cord paralysis compared with patient sex and medical history.

	Vocal cord paralysis		
	Yes (n=34)	No (n=183)	Chi-square *P* value
Female	21 (21)	79 (79)	.046
Male	13 (11)	104 (89)	.046
ICU admission	22 (21)	85 (79)	.051
Intubation	28 (28)	71 (72)	<.001
Cerebral palsy	5 (21)	19 (79)	.460
Gastroesophageal reflux disease	13 (17)	63 (83)	.669

Data are n (%).

Table 5. Association between patient symptoms and signs compared with selected laryngoscopic findings.

	Laryngomalacia (n=149)	Vocal cord paralysis (n=34)	Laryngopharyngeal reflux (n=42)
Weak voice/cry	20/30 (67) .799	14 (47) <.001	5 (17) .688
Positional changes	53 (88) <.001	7 (12) .316	12 (20) .882
Swallowing and feeding difficulties	50 (67) .645	13 (17) .624	21 (28) .019
Failure to thrive	31 (86) .013	6 (17) .857	6 (17) .655
Cough	11 (61) .471	3 (17) .903	3 (17) .763
Cyanosis	19 (79) .240	2 (8) .295	9 (38) .017
Hoarseness	5 (36) .006	9 (64) <.001	3 (21) .839

P* value reported using chi-square. Data in each cell are n (%) and *P* value. Percentages are of the total for the presenting signs and symptoms in **Table 1.

Table 6. Direct laryngobronchoscopy findings (n=39).

Finding	
Subglottic stenosis	19 (49)
Laryngeal cleft	4 (10)
Tracheomalacia	5 (13)
No other findings	13 (33)

Data are n (%).

wheezing (33%), hoarseness (2%), and cyanosis (1%) were the most common symptoms.8 In the current study, weak voice and hoarseness were significantly more likely to be the presenting complaints of patients with vocal cord paralysis. These findings are somewhat in accordance with the results of a similar study that found that vocal cord paralysis was associated with dyspnea (40%), choking and recurrent aspiration (13%), and weak crying (7%).[9] A study by O'Sullivan et al that evaluated 62 patients with stridor with nasopharyngoscopy (NPS) found that 5 of 9 patients with normal NPS had a subglottic lesion on further investigation; this is similar to our finding in that 19 (49%) patients who required further investigation had subglottic stenosis.[6]

The current study has a number of limitations to acknowledge. First, the study sample was collected from a single hospital in Riyadh, which does not reflect the whole population. Second, the study was limited by the small number of patients seen over a period of 37 months. Third, out of all the variables that were collected and compared, only a small number of variables were of statistical significance. However, this study has some strengths overall. First, this is the first study to describe stridor diagnosed with FFL in a Saudi Arabian population. Second, our study used a non-probability consecutive sampling, which included all the patients who fulfilled the inclusion criteria. Thirdly, our study can be used as a comparison for future studies using other evaluation tools including a rigid scope.

In summary, in pediatric patients with stridor, laryngomalacia was the most commonly encountered diagnosis in 149 (69%) patients, followed by laryngopharyngeal reflux in 42 (19%). Furthermore, subglottic stenosis was the most common finding in patients who underwent DLB for further evaluation (n=19, 49%). We conclude that FFL is effective, feasible and easily tolerated by patients as it was sufficient in finding the diagnosis in 178 (82%) patients. The results of this study highlight the variable etiologies of stridor and the significant associated symptoms among different age groups. Moreover, this study demonstrates the effectiveness of a flexible scope in identifying the underlying cause. We recommend further epidemiological and descriptive studies in different regions of Saudi Arabia with the implementation of educational campaigns relating to the importance of investigating patients with stridor.

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