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Bronchoscopic evaluation of chronic stridor in children aged 0-5 years: A study from a Pediatric Tertiary Care Centre from Eastern India.

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Abstract

Background: Stridor is an abnormal, high-pitched monophonic sound, produced by turbulent airflow in a narrowed airway. Its presence suggests significant obstruction of the large airways at the level of the supraglottis, glottis, subglottis or trachea. Stridor can be acute, subacute or chronic. Acute stridor may be due to croup, epiglottis, bacterial tracheitis, and anaphylaxis or due to a foreign body aspiration. Chronic stridor is mainly due to anatomical deformities. There are few studies in the pediatric population regarding bronchoscopic findings in children presenting with chronic stridor.
Aim: To determine the causes of chronic stridor in children less than 5 years using flexible Fibre-Optic Bronchoscopy (FOB).
Method: This retrospective study was conducted from May 2019 to December 2021; 58 children aged less than five years with chronic stridor had undergone FOB within this period. Stridor has been divided into three groups: Inspiratory, expiratory and biphasic. Patients were analyzed regarding sex distribution, cause of stridor and clinical findings. Radiologic investigation reports were included wherever relevant.
Results: Amongst the 58 children, 61% were boys and 39% were girls. The common symptoms associated with chronic stridor were cough (51.7%), breathlessness (32.2%) and fever (19%). The most common sign associated with stridor was tachypnea (82.7%). 22 patients had inspiratory stridor, 30 had biphasic stridor and 6 had expiratory stridor. Of the 58 children with chronic stridor, 44 (75.8%) had abnormal FOB findings. Laryngomalacia was the most common abnormality (31%) followed by subglottic stenosis (15.5%) and tracheomalacia (12%).
Conclusion: In this study of chronic stridor in children aged less than five years, laryngomalacia was the commonest abnormality found on flexible FOB followed by subglottic stenosis and tracheomalacia.

Keywords: Chronic stridor, Flexible bronchoscopy, Laryngomalacia.

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Key Message

Stridor is a common occurrence in pediatric population and suggests an upper air way obstruction. Determining the exact etiology is crucial to management of stridor. The study aims to highlight the importance of FOB in determining the etiology of chronic stridor and elucidates the common causes of stridor.

Introduction

Stridor is a sign of airway obstruction. Depending on the site of obstruction, it can be of 3 types: Inspiratory, expiratory or biphasic [1]. Inspiratory stridor is due to airway obstruction above the glottis while expiratory stridor indicates obstruction lower down the trachea (subglottis). A biphasic stridor indicates a glottic or subglottic lesion. Stridor may be acute, subacute or chronic depending on the duration of the illness [2]. Acute stridor may be due to croup, epiglottitis or foreign body aspiration [3]. Chronic Stridor may be caused by abnormalities in laryngopharynx, larynx or trachea. Laryngomalacia remains the commonest cause of chronic stridor amongst infants. Other causes may include various congenital and acquired conditions like subglottic stenosis, tracheal stenosis, Gastro-Oesophageal Reflux Disease (GORD), foreign body aspiration, vocal cord paralysis, external tracheal obstruction, laryngeal web, haemangioma and cysts [1-3]. It is essential to determine the appropriate aetiology of stridor in order to start appropriate therapy.

Flexible Fibre-Optic Bronchoscopy (FOB) has an important role as a diagnostic modality to determine the aetiology of stridor. It can diagnose both static anatomic or dynamic obstruction in upper and lower airway of children [4]. Though FOB is not a routine investigation in all children with stridor, if done in chronic cases, it can provide a prompt diagnosis. During evaluation of upper airway to find out the cause of stridor, the lower airway should also be evaluated, as around 68% of the patients with upper airway anomalies have associated lower airway anomalies too [5].

Materials and Methods

To describe the clinical characteristics, FOB findings and correlation of findings with clinical symptoms in patients with chronic stridor. This is a single centre retrospective observational study conducted in the institute of child health, Kolkata, India. All children less than 5 years old with clinical complaints of chronic stridor, admitted in the hospital, were enrolled in the study. Patients were divided into 3 groups according to type of stridor: 1) Inspiratory, 2) Expiratory and 3) Biphasic. FOB was done in all these patients having stridor with or without other respiratory findings like wheezing, respiratory distress and cyanosis. Demographic characteristics, symptoms, physical and FOB findings and presence of associated co-morbidities were obtained from the patients' medical records. All these findings were recorded and the data were analyzed.

FOB was performed by the same pulmonologist with a flexible bronchoscope (Ambu a3 scope). The procedure was explained to the parents of the selected children and consent was taken. Before every procedure, both nasal cavities were examined and the wider nostril was selected for introduction of FOB; 2% lignocaine jelly was used as a topical anesthetic agent and was introduced into the nasal cavity before the procedure. The child was given sedation with Intravenous (IV) ketamine or IV midazolam before initiation of the procedure. All emergency drugs and equipment were kept at the bedside. The child was continuously monitored for heart rate, respiratory rate, Oxygen Saturation (SpO_2) and non-invasive blood pressure during the procedure. Continuous oxygen was delivered *via* nasal cannula in order to keep SpO_2 above 92%. FOB was performed through a trans-nasal approach while the patient was breathing spontaneously and under conscious sedation with the above-mentioned drugs. The anatomy of nose, nasopharynx and larynx was noted. Active movements of the vocal cords were observed. After completing the evaluation of the movement of vocal cord and epiglottis, injection 2% lignocaine was flushed through the port of FOB over the vocal cords as local anaesthesia. Gradually the decreased movement of vocal cord was observed and then the scope was negotiated into the trachea to observe the abnormalities of the tracheo-bronchial tree. In selected patients, Broncho-Alveolar Lavage (BAL) was taken and sent for relevant work up.

Ethical issues

Approval for the study was obtained from the institutional ethics committee for biomedical and health research dated 28.04.2022. Consent for participation in the study was taken from the parents of the patients after explaining the procedure of FOB.

Statistical analysis

Data entry was done in Microsoft excel and was statistically analyzed using SPSS software. Descriptive statistical data like means, medians and standard deviations were calculated for continuous variables.

Results

A total of 58 children less than 5 years old with stridor were included in the study. FOB was done in all these selected subjects. The mean age was 22 months and 13 days with the youngest being 18 days old and the oldest being 5 years old. The age distribution of the study group is shown in Table 1.

Age of patient	Number
Less than 1 year	38
1 to <3 years	13
3-5 years	7

Table 1. Age distribution of study group (n=58).

Amongst these, 35 (61%) were boys and 23 (39%) were girls. The most common symptom associated with stridor was cough which was observed in 30 (51.7%) children followed by breathlessness in 18 (32.2%) and failure to thrive in 11 (19%). There were 3 (5%) children who presented with severe respiratory distress involving peripheral and central cyanosis along with grunting.

On examination, the most common finding associated with stridor was tachypnoea which was observed in 48 (82.7%) cases. Based on the clinical history and examination, 14 (24.1%) children were suspected to have laryngomalacia. Table 2 shows the clinical findings in the 3 major causes of stridor.

Clinical feature	Laryngomalacia	Subglottic stenosis	Tracheomalacia
Tachypnoea	12	2	3
Chest retraction	9	4	3
Failure to thrive	4	2	2
Choking	5	0	1
Cough	5	1	0
Heart murmur	1	0	0
Wheeze	0	0	1
Total no of patients	18 (31%)	9 (15.5%)	7 (12%)

Table 2. Clinical findings in the 3 major causes of chronic stridor.

FOB was done under all aseptic precautions as mentioned above and the observations were analyzed. Of the 58 children with chronic stridor 44 (75.8%) had abnormal FOB findings. Laryngomalacia was the most common abnormality observed in 18 (31%) patients followed by subglottic stenosis in 9 (15.5%) and tracheomalacia in 7 (12%). Other abnormalities included neglected foreign body in the airway, external compression of airway, laryngeal oedema, cyst, GORD, congenital web and dynamic tracheal obstruction. Out of the 14 clinically suspected cases of laryngomalacia, 13 (92.8%) were observed to have laryngomalacia on FOB. Of all these cases, 52 (89.7%) children were managed conservatively while 6

(10.3%) were referred for surgical intervention. These cases involved severe laryngomalacia causing significant respiratory distress, foreign body aspiration, congenital web and airway cyst. There was no adverse event in any of the cases during the procedure.

Discussion

Stridor in the pediatric age group is caused mostly by some obstruction in the upper airway [1]. Sometimes it may not have any local cause but may be of some central nervous system pathology [3,6]. Laryngomalacia is the commonest cause of stridor in the first year of life. It presents typically in the first few weeks after birth, may worsen in the first 4-8 months and usually resolves by 18-24 months of age as the child gains weight [7,8]. It is usually managed conservatively but surgical management is necessary in cases with worsening respiratory distress [5]. However, secondary airway lesions other than laryngomalacia may also be present in an insignificant percentage of children where FOB plays an important role in diagnosis [8].

Various studies were performed worldwide to evaluate the cause of stridor in this age group by bronchoscopic evaluation [8-10]. Most of the studies had laryngomalacia as the most common cause of stridor with an associated secondary lower airway anomaly. Laryngomalacia may be due to multiple reasons. One of the most common causes of laryngomalacia is neurological, *i.e.,* altered laryngeal tone due to abnormality of the laryngeal nerve [6,11]. Reflux is another important cause of irritation and oedema of the supra-glottic airway which worsens airway obstruction [12]. Laryngomalacia is mainly a clinical diagnosis having stridor which worsens with feeding and in supine position and decreases on prone positioning [13]. Patients may also have weight loss or failure to thrive. Infants with laryngomalacia are observed to have shortened aryepiglottic folds, floppy omega shaped epiglottis and redundant arytenoid tissue which prolapses over the glottis every time the child breathes [7,8]. In our study, there was a good correlation (92.8%) between clinical diagnosis and bronchoscopic findings in cases with suspicion of laryngomalacia.

There may be other causes of stridor beyond laryngomalacia. In a study by Martins, et al. [14] infra-glottic stenosis, laryngeal inflammation and vocal cord paralysis were the most common findings on bronchoscopy in children with stridor. In a study by Erdem, et al. [2] laryngomalacia was the most common cause for stridor followed by causes like subglottic stenosis, tracheal compression, laryngeal cyst which comprised of 16.5% of the cases evaluated for stridor. In our study, laryngomalacia was the most common cause of stridor (31%) followed by subglottic stenosis (15.5%) and tracheomalacia (12%). Other findings included subglottic stenosis, tracheomalacia, foreign body aspiration, external airway compression etc. Subglottic stenosis involves narrowing of the airway encircled by the cricoid cartilage, below the glottis and above the first tracheal ring [15]. It could be congenital or acquired.

More than 90% of the acquired subglottic stenosis is secondary to endotracheal intubation [16]. Foreign body aspiration is a very common cause of acute onset noisy breathing in the pediatric age group [17]. Classic history of acute onset choking or coughing might not be present in such cases [17,18]. In the study done by Erdem, et al. 15% of the children with stridor required surgical intervention as compared to 10.3% of the children requiring surgical intervention in our study. Stridor improved with time in all cases as observed on follow up. Children with an underlying neurological ailment tend to have persistent stridor for a relatively longer duration [3,7]. However, Yuen, et al. [19] reported that neurological abnormalities did not alter the time to resolution of symptoms.

Bronchoscopic evaluation is very important to accurately diagnose the cause of chronic stridor so that timely and prompt treatment can be initiated according to the aetiology. Pediatric bronchoscopy can be done very easily and without any adverse event by a trained pulmonologist, intensivist, surgeon or anesthesiologist and should be included among the primary investigations in evaluation of children with chronic stridor.

Conclusion

In this study of chronic stridor in children aged less than five years, laryngomalacia was the commonest abnormality found on flexible FOB followed by subglottic stenosis and tracheomalacia. There was good correlation (92.8%) between clinical diagnosis and bronchoscopic findings in cases with a suspicion of laryngomalacia. In the study 15% of the children with stridor required surgical intervention, while in our study, 10.3% of the children needed surgery. Following up revealed that in every case, the stridor got better over time. Children who have a neurological condition at the root of their stridor likely to experience it for a longer period of time. A paediatric bronchoscopy should be among the initial investigations in the evaluation of children with chronic stridor since it can be performed simply and safely by a qualified pulmonologist, intensivist, surgeon, or anesthesiologist.

Ethical Clearance

Approval for the study was obtained from the institutional ethics committee for biomedical and health research of the institute of child health, Kolkata, India (No. ICH/IECBMHR/12/2022) dated 28.04.2022. Consent for participation in the study was taken from the parents of the patients after explaining the procedure of FOB.

Author Contribution

Authors Apurba Ghosh, Ritabrata Kundu and Partha Pratim Halder contributed to conception and design of the study. Partha Pratim Halder, Jigna Bathia, Aniruddha Ghosh and Subhajit Dey Sarkar contributed to acquisition of data and analysis and interpretation of data. Partha Pratim Halder, Jigna Bathia drafted the article and it was revised by Aniruddha Ghosh, Apurba Ghosh and Ritabrata Kundu. Final approval of the version was permitted by Apurba Ghosh and Ritabrata Kundu.

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