Evaluation of Etiology in Infants with Recurrent Pneumonia and Chronic Stridor

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OBJECTIVES: Stridor is a sign of upper airway obstruction. In children, laryngomalacia is the most common cause of chronic stridor, while croup is the most common cause of acute stridor. Stridor may be inspiratory, expiratory, or biphasic. In this study, we aimed to present clinical features and treatments of patients admitted with chronic stridor and recurrent pulmonary infections.

MATERIAL AND METHODS: Twelve patients hospitalized because of chronic stridor and recurrent pneumonia were analyzed clinically, radiologically and with regard to treatment methods. Early onset, frequent recurrence, and persistence of the symptoms, poor improvement in coexistent respiratory distress inspite of appropriate treatment, and also the presence of stridor pushed us to further evaluation. Patients presenting with acute stridor and mild laryngomalacia were excluded from the study.

RESULTS: Five patients were diagnosed with vascular ring, whereas other seven patients presented with tracheomalacia, severe laryngomalacia, laryngeal hamartoma, cervical bronchogenic cyst, foreign body aspiration, H-type tracheoesophageal fistula, and congenital subglottic stenosis.

CONCLUSION: Noisy breathing that begins in the newborn and infancy periods, recurrent lower respiratory tract infections and associated symptoms of airway obstruction should suggest structural and functional abnormalities of the lungs and airways. In patients with recurrent respiratory problems accompanied by stridor, diagnosis can be made with comprehensive physical examination and radiological methods.

KEY WORDS: Airway obstruction, congenital abnormalities, stridor, infant

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INTRODUCTION

Stridor is a high-pitched sound resulting from turbulent air flow that occurs due to partial upper airway obstruction. If upper airway obstruction is above the glottis, it suggests an inspiratory stridor. If it is in the lower trachea, it suggests an expiratory stridor. A biphasic stridor develops in glottic or subglottic lesions [1,2]. In children, the most common cause of acute stridor is croup and the most common cause of chronic stridor is laryngomalacia [3,4].

Recurrent pulmonary infection is defined as having two or more pulmonary infection attacks in a single year or three or more attacks in any time period. Recurrent pulmonary infections can occur due to congenital anomalies, impaired airway secretion clearance, aspiration syndromes, and allergic and immunological reasons [5].

In this study, it was aimed to present clinical, radiological, and treatment features of the patients admitted with chronic stridor and recurrent lower respiratory tract infection.

MATERIAL AND METHODS

Twelve patients hospitalized owing to the diagnosis of chronic stridor and recurrent pulmonary infection in the infancy unit of Dr. Sami Ulus Obstetrics, Children Health and Diseases Education and Research Hospital, Ankara, Turkey, between the dates of January 2006 and January 2008, were evaluated clinically, radiologically, and with regard to treatment methods.

Some analyses were performed for recurrent pulmonary infection considering medical background and physical findings of the patients. All of the patients were assessed with echocardiography (EKO), and with sweat and tuberculin tests. Their immunoglobulin (Ig) M, A, G, E levels were examined.

For radiological evaluation, bilateral lung X-ray graphy, esophagography, neck and thorax computed tomography (CT), and thorax magnetic resonance (MR) angiography were used.
Laryngoscopy was applied to the patients for whom laryngeal pathology was considered. Rigid bronchoscopy was conducted in order to diagnose and treat the patients having been suspected of foreign body. Flexible bronchoscopy was applied for the patients having recurrent pulmonary infection and stridor with unknown etiology. Written informed consent was obtained from the parents of the patients participated in the study. Moreover, ethics committee approval was received from the Ethics Committee of Keçiören Training and Research Hospital, Ankara, Turkey on the date of August 19, 2013.

**Statistical Analysis**

For the categorical variables, the values of number and percentage were used. In addition, standard deviation (SD) values were employed as a measure of variability when the means were given.

**RESULTS**

The mean age of the evaluated cases was 5.1±2.7 months SD (2-8 months) and the ratio of male/female (7/5) was found to be 1.4. All patients were hospitalized in order to treat acute community-acquired pneumonia and to evaluate the etiology of recurrent pulmonary infection and chronic stridor.

Early onset of patients’ complaints, recurrent pneumonia symptoms with respiratory distress despite providing appropriate treatment and the presence of associated chronic stridor led to further examination. Five patients were diagnosed with vascular ring and other patients were diagnosed with tracheomalacia, severe laryngomalacia, cervical hamartoma, cervical bronchogenic cyst, foreign body aspiration, H-type tracheoesophageal fistula (TEF), and congenital subglottic stenosis. Demographic, clinical, laboratory, radiological and bronchoscopic findings of the patients and also treatment methods are shown in Table 1.

Of the patients with chronic stridor who were involved in the study, 85% had persistent stridor and 15% had recurrent stridor. Seven patients (58%) had the complaints beginning from the birth, whereas the complaints of 5 patients (42%) became clear in the first or second months. The complaints of 3 patients (25%) were related to position and they decreased in lying position. Feeding difficulty and malnutrition were found in six patients (50%).

Chronic stridor and accompanying cardiac anomalies in the patients with recurrent pulmonary infection are demonstrated in Table 1. Sweat test results and Ig M, A, G, E levels were found to be normal for the patients having the history of recurrent pulmonary infection. On the other hand, the result of tuberculin test, which was applied to all patients, was positive for one case. Tuberculosis was excluded with the examinations for this patient.

Of the cases evaluated, vascular ring was the most frequently diagnosed defect (42%). Common characteristic of these cases was that symptoms were not associated with position and/or nutrition. In the esophagography conducted with the pre-diagnosis of vascular ring, various indentation patterns were observed in all cases (Figure 1). The final diagnosis was made by defining vascular pathology via MR angiography.

In a case with recurrent upper and lower respiratory tract symptoms and malnutrition, flexible laryngoscopy results were found to be consistent with laryngomalacia results. For this patient with the history of developmental delay and recurrent lower respiratory tract infection, flexible bronchoscopy was performed to evaluate accompanying tracheobronchial pathology and the result was found to be normal.

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**Table 1. Clinical, laboratory, radiological, bronchoscopic and treatment features of the patients**

<table>
<thead>
<tr>
<th>Diagnosis</th>
<th>Age of initial complaint (month)</th>
<th>Age of diagnosis (month)</th>
<th>Stridor</th>
<th>Esophagography</th>
<th>PPD (mm)</th>
<th>ECHO</th>
<th>Bronchoscopy (type)</th>
<th>Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>Aberrant right subclavian artery*</td>
<td>5</td>
<td>congenital</td>
<td>inspiratory</td>
<td>indentation</td>
<td>0</td>
<td>No right subclavian exit</td>
<td>not performed</td>
<td>Follow-up</td>
</tr>
<tr>
<td>Severe laryngomalacia</td>
<td>2</td>
<td>congenital</td>
<td>inspiratory</td>
<td>not performed</td>
<td>0</td>
<td>N</td>
<td>laryngomalacia (flexible)</td>
<td>surgery</td>
</tr>
<tr>
<td>Double aortic arch*</td>
<td>4</td>
<td>2-month</td>
<td>biphasic</td>
<td>indentation</td>
<td>2</td>
<td>ASD</td>
<td>pulsation (flexible)</td>
<td>surgery</td>
</tr>
<tr>
<td>Double aortic arch*</td>
<td>2</td>
<td>congenital</td>
<td>inspiratory</td>
<td>not performed</td>
<td>0</td>
<td>N</td>
<td>not performed</td>
<td>surgery</td>
</tr>
<tr>
<td>H-type TEF</td>
<td>6</td>
<td>congenital</td>
<td>inspiratory</td>
<td>fistule</td>
<td>0</td>
<td>N</td>
<td>not performed</td>
<td>surgery</td>
</tr>
<tr>
<td>Right aortic arch accompanied by ligamentum arteriosum*</td>
<td>5</td>
<td>congenital</td>
<td>inspiratory</td>
<td>indentation</td>
<td>1</td>
<td>VSD</td>
<td>not performed</td>
<td>surgery</td>
</tr>
<tr>
<td>Cervical bronchogenic cyst</td>
<td>4</td>
<td>congenital</td>
<td>inspiratory</td>
<td>not performed</td>
<td>12</td>
<td>N</td>
<td>not performed</td>
<td>surgery</td>
</tr>
<tr>
<td>Cervical hamartoma</td>
<td>7</td>
<td>3-month</td>
<td>inspiratory</td>
<td>not performed</td>
<td>0</td>
<td>VSD</td>
<td>not performed</td>
<td>surgery</td>
</tr>
<tr>
<td>Subglottic stenosis</td>
<td>6</td>
<td>congenital</td>
<td>biphasic</td>
<td>not performed</td>
<td>0</td>
<td>N</td>
<td>not performed</td>
<td>surgery</td>
</tr>
<tr>
<td>Tracheomalacia</td>
<td>7</td>
<td>1-month</td>
<td>expiratory</td>
<td>N</td>
<td>3</td>
<td>N</td>
<td>Tracheomalacia (flexible)</td>
<td>Follow-up</td>
</tr>
<tr>
<td>Vascular sling</td>
<td>7</td>
<td>congenital</td>
<td>biphasic</td>
<td>indentation</td>
<td>0</td>
<td>Silent ductus</td>
<td>N (flexible)</td>
<td>surgery</td>
</tr>
<tr>
<td>Foreign body aspiration</td>
<td>6</td>
<td>4-month</td>
<td>inspiratory</td>
<td>not performed</td>
<td>0</td>
<td>N</td>
<td>Foreign body (rigid)</td>
<td>Removal of foreign body</td>
</tr>
</tbody>
</table>

ECHO: echocardiography; ASD: atrial septal defect; VSD: ventricular septal defect; TEF: tracheoesophageal fistula; N: normal

*Vascular Ring
In the case without a history of aspiration, who had had respiratory tract symptoms for two months, foreign body aspiration was suspected because of the inequalities in pulmonary auscultation results and the difference in ventilation revealed by direct radiography. The hull of sunflower seed was removed with rigid bronchoscopy.

In another patient presented with the complaints of failure to thrive, stertorous respiration since birth, and recurrent lower respiratory tract infection, the diagnosis of cervical bronchogenic cyst was made after long-lasting mechanic ventilation monitoring. The location of lesion was defined with cervical CT (Figure 2). Malnutrition and respiratory tract complaints of the operated patient disappeared completely.

In a case with recurrent respiratory tract infection and persistent progressive stridor, surgical excision was performed for removing the cervical lesion which was determined via laryngoscopy and cervical CT. Clinical findings of the patient who was diagnosed with laryngeal hamartoma depending on the pathology results improved.

**DISCUSSION**

Since the reflex mechanism providing larynx to open and close is insufficient in infants, tendency to aspiration and laryngospasm increases, which may result in obstruction. In children, subglottic region is the narrowest part of the airway, so even a few millimeter mucus or edema may cause life threatening airway stenosis [3,5,6]. Therefore, the patients admitted with the finding of stridor should be classified as acute or chronic at the beginning. The patients with chronic stridor should be evaluated taking persistent or recurrent characteristic of the disease into consideration [7,8]. The diseases that should be considered for the patients presented with stridor are shown in Table 2.

Stridor may be classified as inspiratory, expiratory, or biphasic depending on the location of obstruction. Because the support of supraglottic region is poor, pathologies in this region lead to inward collapse and high-frequency inspiratory stridor due to the negative pressure over the air passage occurred in inspirium. Changes in lumen pressure do not affect diameter since cartilage support of glottic or subglottic regions is stronger, and both inspiratory and expiratory occur in narrowing in these regions [9]. Inspiratory stridor often occurs with pharyngeal, laryngeal and nasal lesions out of the thorax, while expiratory stridor occurs with tracheal and bronchial lesions in the thorax [2,3,10]. Of the cases involved in the study, 8 patients (67%) had inspiratory stridor, 3 (25%) had biphasic stridor, and 1 (8%) had expiratory stridor.

Medical history and physical examination findings are used for the differential diagnosis of stridor. Also, the onset and time of stridor are important for the differential diagnosis. Stridor congenital anomalies since birth suggest stridor laryngomalacia and tracheomalacia that begin in 4-6 weeks while acute stridor beginning at the age of 1-4 years often suggest infections or foreign body aspiration [3,6,11]. In the study, it was found that 7 patients (58%) had symptoms beginning from the birth and the symptoms of 5 patients (42%) began in the first or second month after birth and continued chronically.

**Table 2. Causes of chronic stridor**

<table>
<thead>
<tr>
<th>Causes of persistent stridor</th>
<th>Causes of recurrent stridor</th>
<th>Other causes of stridor</th>
</tr>
</thead>
<tbody>
<tr>
<td>Laryngeal obstruction</td>
<td>Extrinsic masses</td>
<td>Tracheobronchial diseases</td>
</tr>
<tr>
<td>• Laryngomalacia</td>
<td>• Mediastinal masses</td>
<td>• Tracheomalacia</td>
</tr>
<tr>
<td>• Papilloma and other tumors</td>
<td>• Vascular ring</td>
<td>• Subglottic tracheal weber</td>
</tr>
<tr>
<td>• Laryngocele and cysts</td>
<td>• Lobar emphysema</td>
<td>• Endotracheal, endobronchial tumors</td>
</tr>
<tr>
<td>• Laryngeal web</td>
<td>• Bronchogenic cysts</td>
<td>• Subglottic tracheal stenosis</td>
</tr>
<tr>
<td>• Vocal cord paralysis</td>
<td>• Thyroid compression</td>
<td>• Tracheoesophageal fistula</td>
</tr>
<tr>
<td>• Foreign body</td>
<td>• Foreign body in esophagus</td>
<td></td>
</tr>
<tr>
<td>• Laryngeal cleft</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Infections (Laryngitis, Tracheitis, Epiglottitis)</td>
<td>Allergic (spasmodic) croup</td>
<td>Laryngomalacia</td>
</tr>
<tr>
<td>Gastroesophageal reflux</td>
<td>Macroglossia</td>
<td>Hysterical stridor</td>
</tr>
<tr>
<td>Cri-du-chat Syndrome</td>
<td>Pierre Robin Syndrome</td>
<td>Hypocalcemia</td>
</tr>
<tr>
<td></td>
<td></td>
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</tr>
</tbody>
</table>

In the case without a history of aspiration, who had had respiratory tract symptoms for two months, foreign body aspiration was suspected because of the inequalities in pulmonary auscultation results and the difference in ventilation revealed by direct radiography. The hull of sunflower seed was removed with rigid bronchoscopy.

In another patient presented with the complaints of failure to thrive, stertorous respiration since birth, and recurrent lower respiratory tract infection, the diagnosis of cervical bronchogenic cyst was made after long-lasting mechanic ventilation monitoring. The location of lesion was defined with cervical CT (Figure 2). Malnutrition and respiratory tract complaints of the operated patient disappeared completely.

In a case with recurrent respiratory tract infection and persistent progressive stridor, surgical excision was performed for removing the cervical lesion which was determined via laryngoscopy and cervical CT. Clinical findings of the patient who was diagnosed with laryngeal hamartoma depending on the pathology results improved.
Laryngomalacia is the most frequent cause of chronic stridor in the children younger than the age of 2 years. Inspiratory stridor, that increases with upper respiratory tract infections, crying, discomfort and lying position, is observed [9,15]. Although surgical treatment is not needed most of the time, our patient with severe laryngomalacia was operated due to severe malnutrition and recurrent respiratory tract infection.

Foreign body aspiration is a common cause of acute stridor. However, the history of foreign body aspiration is not clear in some situations. Acute clinical picture is not recognized and patients present with recurrent respiratory tract infection [1,16,17]. In a six-month patient with stridor lasting for two months, who was hospitalized twice because of pneumonia, although there was no history of foreign body aspiration, a foreign body was identified with rigid bronchoscopy and removed.

Vascular rings are categorized as complete vascular rings surrounding trachea and oesophagus (right aortic arch accompanied by double aortic arch and ligamentum arteriosum), and incomplete vascular rings compressing trachea and oesophagus (aberrant right subclavian artery, innominate artery and vascular sling) [18-20]. The diagnosis of vascular ring is made through CT and MR angiography after monitoring of indentation with esophagography [19,21]. The treatment of vascular ring is surgical and symptoms often disappear with treatment. Asymptomatic aberrant right subclavian artery is treated without surgical operation. On the other hand, for vascular sling, symptoms may not disappear even after operation [22,23]. In the study, the patient with aberrant right subclavian artery was monitored due to the mild course of pressure findings in spite of its being symptomatic. Other patients with vascular ring were operated and the complaints of all patients, except the one with vascular sling, disappeared after operation.

Bronchogenic cysts arising with abnormal development of the tracheobronchial tree during the embryological period appear mostly in the mediastinum and lung parenchyma. In the region of neck, thyroglossal, thymic and bronchial cleft cysts are more common, but bronchogenic cysts are rarely seen [24,25]. In the study, cervical bronchogenic cyst was determined and surgical excision was performed in the 4-month patient having congenital respiratory tract symptoms with progression.

Hamartomas are nonneoplastic developmental anomalies which are uncommon. They appear more frequently in the lungs and nasopharynx, but rarely in larynx [26]. In the study, rarely seen cervical hamartoma found in a patient was removed with surgery and the complaints of the patients disappeared completely.

Tracheomalacia can be congenital or acquired. It is characterized with the abnormal collapse of cartilaginous and myoelastic supporting components of the trachea. Congenital tracheomalacia results from incomplete maturation of tracheobronchial cartilages. It may not be apparent at birth and it becomes evident in the first weeks of life. The diagnosis of tracheomalacia is made through dynamic imaging of the trachea and bronchus, which shows airway collapse during
Exhalation [27,28]. In the study, one of the patients had persistent expiratory stridor and recurrent pulmonary infection beginning from the age of one month and he/she was followed with the diagnosis of infantile asthma until the age of 7 months. For the patient, all results had been found to be normal in the previous examinations. Flexible bronchoscopy was performed for this patient and he/she was diagnosed with tracheomalacia. As seen in this case, if the cause of recurrent pulmonary infection and stridor cannot be identified in a patient, flexible bronchoscopy should be performed definitely.

Esophageal atresia and TEF are defined as the ongoing fistulous relationship between the esophagus and trachea due to occurrence of a discontinuation during separation period. Normally, they would form as separate tubes during embryological development. It has five different anatomic types. The frequency rate of H-type TEF is 4%. H-type TEF should be suspected if cyanosis, coughing and stridor attacks occur with feeding. It is diagnosed through esophagography [4,29]. In the study, cyanosis and stridor attacks were observed in a 6-month patient admitted with recurrent lower respiratory tract infection while feeding and the diagnosis of H-type TEF was made through barium esophagography. The patient was operated by a pediatric surgeon. His/her complaints disappeared and then he/she was monitored.

Congenital subglottic stenosis is one of common congenital larynx anomalies. It results from inadequate recanalization after epithelial fusion during embryological development. It may be asymptomatic or appear with the symptom of dysphagia and feeding difficulty. It can lead to airway obstruction by causing an additional narrowing in upper respiratory tract infections. Also, biphasic stridor, dyspnoea, suprasternal and supraclavicular retraction appear [30]. In the study, severe biphatic stridor increasing with upper respiratory tract infections was identified in a patient with the diagnosis of congenital subglottic stenosis. The patient was admitted with severe stridor as an emergency case and subsequently intubated due to respiratory arrest. During intubation, the smallest endotracheal tube was hardly passed. The patient evaluated by the otolaryngologists underwent operation urgently for tracheostomy. During the operation, congenital subglottic stenosis was determined, thus stenosis surgery was performed instead of tracheostomy. In such cases, flexible bronchoscopy can be used as well as patient’s medical history, physical examination, and radiological findings for diagnosis.

Although the patients in the study presented with similar clinical findings, detailed information about the diseases was limited due to the diversity of the diagnoses.

Consequently, if the findings of noisy breathing that begins in the newborn and infancy periods, recurrent lower respiratory tract infection and associated airway obstruction are present, structural and functional anomalies of the lungs and airways should be taken into consideration. Since positive outcomes can be obtained through early surgical treatment in structural airway anomalies, diagnosis should be made early. Increased stridor and respiratory distress related to nutrition and position should suggest vascular rings. In these cases, determination of indentation through esophagography is an important guide before CT or MR angiography that will be performed for the final diagnosis. In the patients with recurrent respiratory problems accompanied by stridor, diagnosis can be made with a comprehensive medical history, physical examination, and radiological methods. Flexible bronchoscopy should be absolutely performed for all patients admitted with recurrent pulmonary infection and stridor with unknown reason.

Ethics Committee Approval: Ethics committee approval was received for this study from the ethics committee of Keçiören Training and Research Hospital, Ankara, Turkey. (08.19.2013).

Informed Consent: Written informed consent was obtained from the parents of the patients who participated in this study.

Peer-review: Externally peer-reviewed.


Conflict of Interest: No conflict of interest was declared by the authors.

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REFERENCES


25. Maung KH, Low C, Knight LC, Cullinane Cl. Multiple cervical bronchogenic cysts. J Laryngol Otol 2006;120:145-7. [CrossRef]


