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Case Report

A Case Presented with Tracheal Compression Caused by An Anatomical Anomaly: The Innominate Artery Syndrome

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Abstract

Innominate artery compression syndrome is a rare congenital anomaly. The condition is an important consideration in the differential diagnosis of patients presenting with noisy breathing, a barky cough, and expiratory stridor. Here we report a case of Innominate Artery syndrome that presented with persistent and /or biphasic stridor in a 3-month-old. This case provides us to highlights the importance of unequivocally identifying the vascular anomalies and to emphasize once again the importance of detailed history and observing/hearing of the breathing during the examination.

Introduction

Innominate artery compression syndrome (IACS) or named as brachiocephalic trunk compression syndrome extremely rare congenital vascular anomaly that classically characterized by a significant compression of the anterior portion of the trachea by the artery as it crosses from left to right after originating from the ascending thoracic aorta. The anomaly first described by Gross and Neuhauser in 1948 [1,2]. It generally presents with noisy breathing, a barky cough, expiratory stridor, recurrent cough, apnea, wheezing, dyspnea on exertion, dysphagia, reflex apnea or dying spells, increased work of breathing, and recurrent bronchopulmonary infection. Chest magnetic resonance images, computed tomographic angiograms, and rigid bronchoscopy are usually utilized for its diagnosis [2,3]. Anterior compression of the trachea may be demonstrated on airway fluoroscopy or rigid bronchoscopy that typically shows a compression as short segment, located a few centimeters above the carina [4].

Case Report

A 3-month-old boy was referred to our clinic with a 2-months history of noisy breathing, a barky cough, and expiratory stridor. His birth weight was 3,470 g and gestational age was 39 weeks. Apgar score was 8 and 9 at 1 and 5 minutes, respectively. The pregnancy was complicated by polyhydramnios. The diagnosis of esophageal atresia was confirmed after the failure to pass a nasogastric tube. This was checked by an esophagogram, which also showed a distal tracheoesophageal fistula. Post-operation 43rd day, before admission to our clinic, he developed noisy breathing, a barky cough, biphasic stridor, and cyanosis during crying and feeding. It was found out that the patient was mostly diagnosed with "trachea-laryngomalacia" due to expiratory stridor, and barky cough; he was evaluated with airway fluoroscopy for diagnosis of tracheal anomalies, and there were no pathologies associated with the larynx, glottic area, and tracheal anomalies. On admission, when we approached the patient, expiratory stridor, noisy breathing, a barky cough were clearly heard. Additionally, he was conscious but was suffered from increased work of breathing and during feeding and crying; severe cyanosis with reflex apnea or dying spells were clearly observed. Following his initial hospitalization, he continued to be a very noisy breather. It was described episodes of gasping with loss of breath. These apnea-like episodes were at times associated with feeding and crying. On physical examination, his vital signs were normal for age, respirations were 28 breaths per minute. SpO2 was 97% in room air. The height and weight were both near the 25th percentile. No localized wheezes or rhonchi were heard. Subcostal retractions were present during quiet breathing and increased during crying and agitation. Routine blood counts and biochemical investigations were normal. An over again bedside flexible nasolaryngoscopy examination revealed normal glottic and supraglottic structures. The chest X-ray showed mild hyperinflated lung fields bilaterally. Additionally, cervical and proximal thoracic trachea were in normal calibration. A barky cough, expiratory stridor, cyanosis during feeding and crying and after observed reflex apnea, considering the patient's history, it was decided to get a thoracic computed anjio tomography of the lung and cervical area. The anjio computed tomography with 3D reconstruction revealed a compression syndrome, consisting of an Innominate artery compression syndrome. After its origin, the brachiocephalic artery compressed the front of the trachea resulting in a diameter decrease as 3 mm, and there was approximately 50% stenosis in the trachea. The trachea diameter was 6.5 mm during compression and 6.1 mm after compression. The tomography images of this patient were shown in (Figure 1, 2). The echocardiogram did not show other congenital heart problems. He was treated with aortopexia and discharged.

Discussion

In this case, a 3-month-old boy with a history of barky cough and biphasic stridor was presented and diagnosed with an Innominate artery compression syndrome which is a very rare vascular anomaly that is known as the most common cause of vascular airway compression. Innominate artery compression syndrome or known as brachiocephalic trunk compression syndrome is a congenital vascular anomaly resulting from the more distal attachment of an innominate artery along the aortic arch, compressing the respiratory tract as it crosses anteriorly. The tracheal compression reduces with age and that the condition is rarely diagnosed beyond infancy. Although there are several reports of patients whose anomaly is diagnosed later in life, the condition is generally detected in the first year of life [3]. The incidence of a common trunk has been reported varies widely from around 1% to as high as 27%.[4,5]. Most patients with an innominate artery compression syndrome present symptom of noisy breathing a barky cough, expiratory stridor, recurrent cough, apnea, wheezing, dyspnea on exertion, dysphagia. It has also been reported to be associated with other respiratory problems reflex apnea or dying spells, increased work of breathing, and recurrent

bronchopulmonary infection. Bronchial secretion is generally increased which might lead to a vicious circle associated with long-lasting infections and breathing difficulty. As a result of compressing the trachea related to the vascular anomaly; continuous, brassy, staccato-like cough can be seen in these patients. the breathing difficulty and reflex apnea are aggravated during feeding [6]. The presumptive diagnosis may usually be reached via a detailed patient's history and the breathing of the patient should listen during feeding and crying. A story of noisy breathing and a barky cough can often be taken from parents. Patients may present with persistent brassy, staccato-like cough. A chest radiograph should be the first diagnostic study that frequently showed abnormalities including atelectasis, pneumonic infiltrates, and hyperaeration. Nowadays, angiographic studies or Magnetic resonance imaging (MRI) is preferred to confirm that tracheal compression was indeed secondary to the innominate artery. In addition, these radiological methods also help the planning of surgical management. Another route to diagnosis is flexible bronchoscopy that can show extrinsic compression of the trachea [7]. Most patients have a complete recovery of symptoms after surgical intervention. Successful treatment by aortopexy of the artery to the dorsal surface of the sternum has been reported with success rates in 56% to 87% of patients [2].

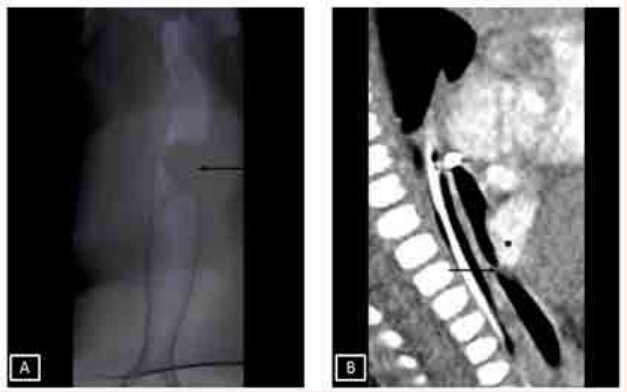

Figure 1A: 3-D and volumetric computed tomography image shows a marked print from the right part of the trachea (black arrow). **1B**: Sagittal CT image shows depression of the trachea anteriorly (black arrow) by the brachiocephalic artery (black star).

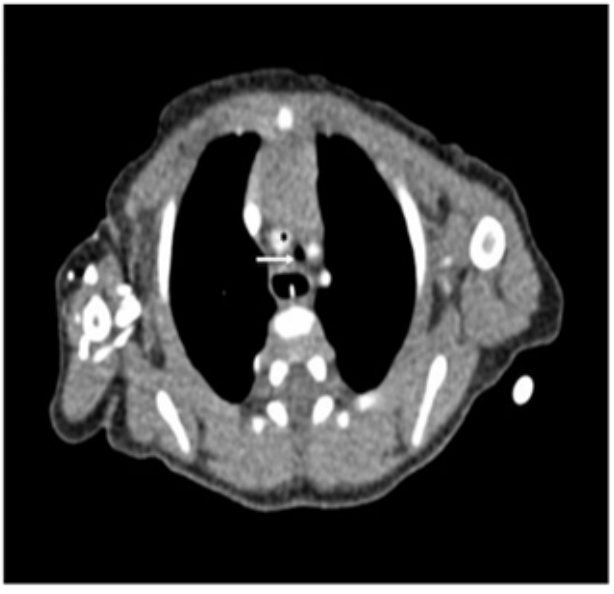

Figure 2: There is compression on the trachea (white arrow) by the brachiocephalic artery (black star) of which the course of to the right hemithorax.

Conclusion

This case highlights the importance of unequivocally identifying the vascular anomalies through diagnostic technique; when an infant or child presents with persistent and /or biphasic stridor, persistent staccato cough, noisy breathing, wheezing, and was unresponsive to treatment; congenital vascular anomalies should be considered, and the diagnostic evaluation should not be delayed. We aimed to call attention to the vascular anomalies, to emphasize once again the importance of detailed history and observing/hearing of the breathing during the examination.

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