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Case Report

Paediatrics Section

Tracheal Compression by Aberrant Innominate Artery: A Case Report

ANKIT AGARWAL[1], NIDHI BEDI[2], PANKAJ ABROL[3], ANKIT PARAKH[4], RENU SHARMA[5]

ABSTRACT

The brachiocephalic trunk arises from the convexity of the aortic arch, posterior to the manubrium sterni. In rare cases, the origin of the brachiocephalic trunk is shifted to the left of the midline, crosses the trachea through an oblique course, and rises upwards. Sometimes this aberrant artery due to an anomalous course can cause a triad of symptoms consisting of cough, stridor, and occasional apnoea known as innominate artery compression syndrome. Here one such case is presented in which, a nine-month-old infant presented with complaints of recurrent cough and noisy breathing since 10 days of age. The infant had a history of frequent hospital admissions for cough since one month of age where he was given nebulisation but with only partial response. A bronchoscopy was done which was suggestive of mid-tracheal compression. Contrast-Enhanced Computed Tomography (CECT) chest confirmed the diagnosis and the child was then referred to a cardiac surgeon for further management. It is important to note that compression of the trachea or tracheobronchial tree by congenital vascular anomalies is not an uncommon cause of stridor and should always be considered a differential diagnosis while evaluating such cases.

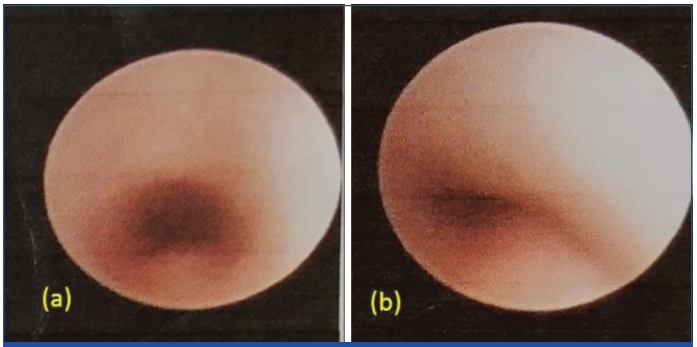
Keywords: Aortic arch syndromes, Brachiocephalic trunk, Tracheal stenosis

CASE REPORT

A nine-month-old infant presented to the paediatric outpatient department with complaints of recurrent cough since 10 days of age. Each episode was sudden in onset, dry in nature, progressively increasing, associated with noisy breathing, and not accompanied with blood and sputum. There were no aggravating or relieving factors or diurnal or seasonal variations. There was no history of fever, poor oral intake or improper weight gain, cyanosis, diarrhoea, rash, weight loss, or abnormal body movements. There was no history of tuberculosis in the family. There was a history of frequent hospital admissions of the child for cough since one month of age where he was given nebulisations with bronchodilators and inhaled corticosteroids but with only partial response each time. The infant was born of non consanguineous marriage, through vaginal delivery with a birth weight of 3.4 kg and cried immediately after birth, with uneventful antenatal, natal, and postnatal periods. He achieved developmental milestones as per age and received all immunisation as per the national immunisation schedule. The child was exclusively breastfed till four months of age followed by mixed feeds. Complementary feeds were initiated at six months of age.

Anthropometric parameters were within normal limits {weight 8 kg (0 to -1SD), length 70 cm (0 to -1SD) and head circumference 46 cm (0 to +1SD)}. At presentation in the present episode, the child was tachypoenic with a respiratory rate of 42/min pulse rate of 116/min, blood pressure of 88/60 mmHg, temperature of 98.2° F and SpO_2 of 98% on room air. General physical examination did not reveal any abnormal findings. Respiratory system examination showed centrally placed trachea and chest wall shape was symmetric with equal, symmetric movements on both sides. Breath sounds were bilaterally equal with vesicular character with bilateral wheeze. The rest of the systemic examination including the cardiovascular system was normal. Laboratory reports revealed haemoglobin levels of 11.6 g/dL, a total leukocyte count of 10,500/mm^3, and a platelet count of 600,000/mm^3. The C-Reactive Protein (CRP) level was 6.1 mg/L, and the blood culture results were sterile. Both kidney and liver function tests were within normal limits. A chest X-ray showed bilateral infiltrates. Nebulisation with bronchodilators and inhaled corticosteroids (budesonide 250 µg 12 hourly) were started following which initial improvement was noted but subsequently developed worsening of symptoms after 2-3 days. After ruling out infective aetiology, oral steroids were initiated and further work-up was continued.

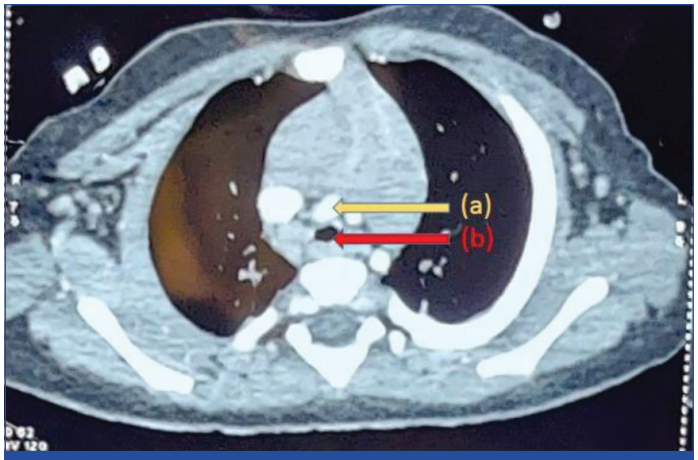
In view of poor response, the possibility of congenital malformations of the respiratory tract, foreign body aspiration and congenital heart disease was kept. A 2D ECHO was done and reported to be normal. Paediatric pulmonology opinion was taken and the child underwent bronchoscopy which was suggestive of mid tracheal compression on the anterior wall [Table/Fig-1]. Subsequently, CECT chest was done to look for the vascular cause of compression and the finding revealed asymmetric narrowing of the tracheal lumen on the right-side in mid tracheal lumen and presence of innominate artery coursing just anterior to this part [Table/Fig-2]. Thus diagnosis of tracheal compression by an aberrant innominate artery was made and the child was referred to a cardiothoracic surgeon for further management.



[Table/Fig-1]: Fiber optic bronchoscopy of a nine-month-old child presented with recurrent cough showing: a) Trachea; b) Compression of trachea on the anterior wall.

DISCUSSION

Vascular ring anomalies can cause significant respiratory and gastrointestinal issues due to compression of the trachea and oesophagus [1]. These pathologies are classified into complete and incomplete vascular rings [2]. Complete ring refers to the abnormal vascular structure or their remnants forming a complete ring around the trachea and oesophagus [3]. The double aortic arch and right



[Table/Fig-2]: Contrast-Enhanced Computed Tomography (CECT) chest of a nine-month-child presented with recurrent cough showing: a) Innominate artery coursing anterior to the trachea; b) Trachea.

aortic arch with left ligamentum arteriosum are included under the complete vascular ring [4]. The case described involves tracheal compression, which aligns with previously reported cases of aberrant innominate artery causing anterior tracheal compression.

The aberrant innominate artery originates more to the left and follows an aberrant route crossing the trachea anteriorly thereby, causing tracheal compression. The spectrum of signs and symptoms varies from mild stridor and/or wheeze to dyspnoea, cough, difficulty in feeding, cyanosis and reflux apnoea [5].

Key findings of this case include the patient's persistent respiratory symptoms, including stridor, wheezing, and recurrent infections, which are indicative of vascular ring pathologies [6].

Fiber-optic bronchoscopy revealed airway compression and subsequent CECT confirmed the diagnosis [7]. The diagnostic approach is consistent with the established literature, supporting the utility of bronchoscopy and imaging in identifying vascular anomalies [8].

What makes this case unique, compared to previous cases, is the complexity of symptoms and the delay in definitive diagnosis. While other reports, such as the study by Wine TM et al., have established surgical management as crucial in cases with recurrent bronchopulmonary infections [9], this case underscores the importance of considering earlier intervention. Surgical indications have evolved over time, with the inclusion of additional factors such as medical management failure, synchronous airway lesions, and failure to thrive, but the delayed diagnosis in this case, led to prolonged patient discomfort. Suspension of the innominate artery to the sternum is the widely accepted treatment [10]. Choice of treatment is based on clinical presentation and degree of tracheal compression, a good clinical outcome may be obtained also in children in whom aortopexy is indicated, that is, those presenting initially with more severe symptoms [11].

This case contributes new insights by demonstrating that, despite the growing body of literature on vascular rings, diagnostic delays remain a challenge. It highlights the need for heightened clinical suspicion in cases with recurrent respiratory symptoms, even when initial management seems adequate. This case advocates for earlier imaging and surgical intervention to prevent complications in similar future cases.

CONCLUSION(S)

Although rare, compression of the trachea or tracheobronchial tree by congenital vascular anomalies is not an uncommon cause of stridor and should always be considered as the differential diagnosis while evaluating such a child. Bronchoscopy should always be considered as part of the investigation while evaluating an infant with recurrent stridor or wheeze.

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PARTICULARS OF CONTRIBUTORS:
1. Junior Resident, Department of Paediatrics, The Faculty of Medicine and Health Sciences, Gurugram, Haryana, India.
2. Professor, Department of Paediatrics, The Faculty of Medicine and Health Sciences, Gurugram, Haryana, India.
3. Professor and Head, Department of Paediatrics, The Faculty of Medicine and Health Sciences, Gurugram, Haryana, India.
4. Consultant Paediatric Pulmonologist, Department of Paediatrics, BLK MAX Hospital, Gurugram, Haryana, India.
5. Senior Resident, Department of Paediatrics, The Faculty of Medicine and Health Sciences, Gurugram, Haryana, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Ankit Agarwal,
Junior Resident, Department of Paediatrics, The Faculty of Medicine and Health Sciences, Gurugram-122505, Haryana, India.
E-mail: agarwal9411@gmail.com

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