

Reduced Exercise Tolerance Does Not Always Mean Bronchial Asthma

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Reduced exercise tolerance in a patient in whom any heart diseases were preliminarily excluded suggests other frequent causes of effort-related symptoms, that is, exercise-induced bronchoconstriction, lack of training, or overweight. The presented case underscores the importance of comprehensive diagnostic workup in children with exercise-related symptoms.

Keywords: reduced exercise tolerance, asthma, spirometry

Introduction

DIFFERENTIAL DIAGNOSTIC OF reduced exercise tolerance in a child should include diverse possible causes. Exercise-induced bronchospasm, exercise-induced asthma, and the lack of training are most commonly reported.^{1,2} Other less frequently observed causes, such as laryngomalacia, inhalation of a foreign body, vocal cord dysfunction, exercise-induced anaphylaxis, and the vascular ring, should also be considered.³

Case Description

A 7.5-year-old boy was referred to the department of pneumology and cystic fibrosis with the suspicion of bronchial asthma. The child had a history of recurrent obstructive bronchitis in infancy, frequent upper respiratory tract infections during the autumn–winter season, often with accompanying hoarseness. Frequency of infections decreased with age. As per parents' report, reduced exercise tolerance, observed from ~3 years, was the dominant complaint.

During the perinatal period, the boy did not show any disorders. To date, his development was normal. In the last 3 years, the boy reported fatigue and the feeling of “being out of breath” or “heavy breathing,” occurring during an intense physical effort (running and playing outdoors). These complaints would often limit the exercise tolerance. No wheezing or coughing after a physical activity was observed and, at that time, no inhaled β_2 agonists were prescribed to treat the complaints. At the age of 6 years, the child was consulted by a cardiologist. Electrocardiography and echocardiography (ECHO) examinations were performed that were both unremarkable. A chest X-ray was not performed.

On admission, the child was in a good condition, with no symptoms of an ongoing respiratory infection (e.g., sneezing, runny nose, and sore throat). Physical examination revealed the following abnormalities: obesity [body mass index 22.04 kg/m², >95–97 percentile (+1SD); body weight 38.4 kg, >97 percentile (+2SD)] and a circumscribed area of hypomelanosis on the skin of the front of the trunk. No other anomalies were observed.

The only abnormalities in laboratory tests were slightly elevated antistreptolysin O value (585 IU/mL) and an increased percentage of neutrophils (62%) in peripheral blood. Skin prick tests were negative. Immunoglobulin levels, quantitative analysis of lymphocyte subpopulations, and the sweat chloride level were all normal. Microbiology tests revealed growth of *Streptococcus pyogenes* in throat swab. Peripheral oxygen saturations measured by pulse oximetry (SpO₂) on admission and during sleep were normal.

The following lung function tests were performed:

- Spirometry showed reduced values of peak expiratory flow (PEF; 62% of the predicted value) and MEF75 (maximal expiratory flow at remaining 75% forced vital capacity; 64% of the predicted value), with normal forced expiratory volume in 1 s (FEV1) (98% of the predicted value), as well as a flattening in the proximal segment of the expiratory flow–volume curve (Fig. 1).
- The inspiratory flow–volume curve was normal (expected to be flattened in upper airways obstruction syndromes)
- Bronchodilation test with a β_2 -agonist was negative (no increase of FEV1 after salbutamol)
- Body plethysmography, similar to spirometry, demonstrated the presence of reduced PEF and MEF75 values (63% and 69%, respectively), and a slightly increased airway resistance (Raw, 132% of the predicted value)

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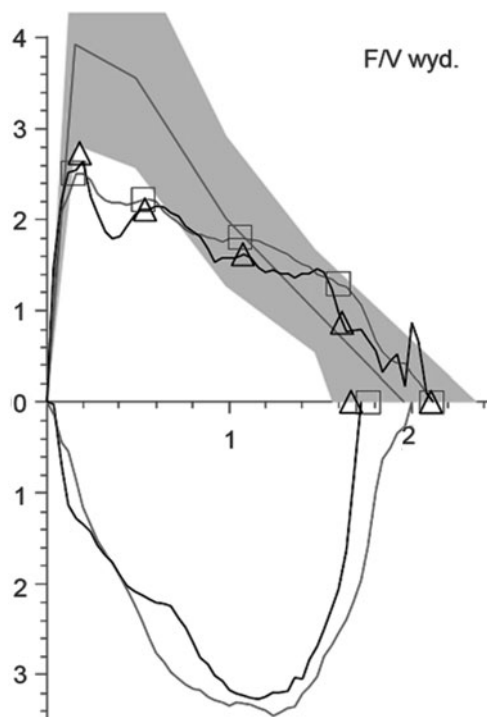


FIG. 1. Spirometry—flattening observed in the proximal segment of the expiratory flow–volume curve (with a normal inspiratory curve), unresponsive to bronchodilator ($\square$ baseline, Δ 15 min. after inhalation of salbutamol).

- Nasal and oral exhaled nitric oxide concentrations were normal
- Diffusing capacity for carbon monoxide was normal (134% of the predicted value).

Considering the reported reduced exercise tolerance, the 6-minute walk test was performed:

- The distance walked by the patient was 657 m (82% of the predicted value)
- After the test, the boy reported mild shortness of breath (2 points according to the Borg CR10 scale⁴), without cough, wheezing, or stridor during or after the exercise
- SpO₂ decreased by 2% from the baseline value (from 99% to 97%)
- A decrease in PEF by $\sim 10\%$ from the baseline value was observed, and an increase in the heart rate value from 86 to 153/min.

Chest X-ray examination revealed a well-saturated shadow in the superior mediastinum, on the right side, merging with the shadow of the spine (Fig. 2). The lateral image of the chest was normal. Computed tomography (CT) of the chest with intravenous contrast revealed right-sided aortic arch, modeling the cross-section of the trachea (Figs. 3A and B).

Fiberoptic bronchoscopy confirmed pulsating compression of the trachea ~ 2 cm above the carina protruding into the right anterolateral wall, corresponding with a large vessel at this site.

Following diagnosis was made based on diagnostic tests: Right-sided aortic arch and obesity.

The patient was discharged home and urgently referred for cardiology consultation. ECHO confirmed right-sided

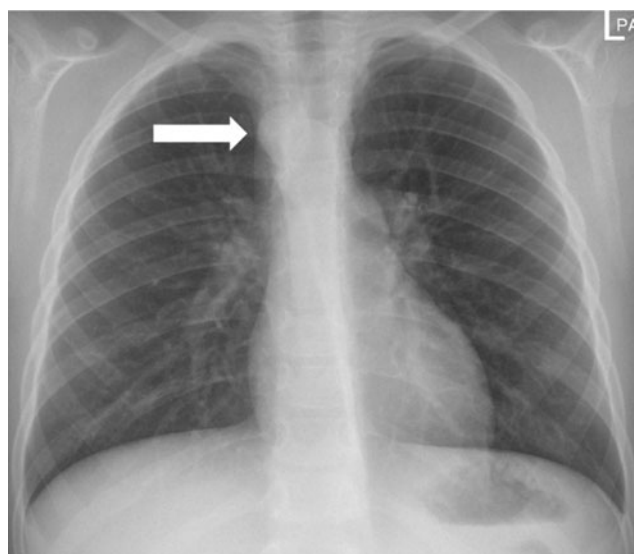


FIG. 2. Chest X-ray of the patient, posteroanterior (PA) projection—a well-saturated shadow observed in the upper mediastinum on the right side (arrow).

aortic arch and revealed the vessel protruding from the arch that might suggest Kommerell's diverticulum, a rare aortic arch anomaly. The boy was qualified for a surgery, which was performed 4 months after diagnosis. Intraoperatively, the right-sided aortic arch and hypoplastic left aortic arch were found. Besides, a filled blood vessel was discovered,

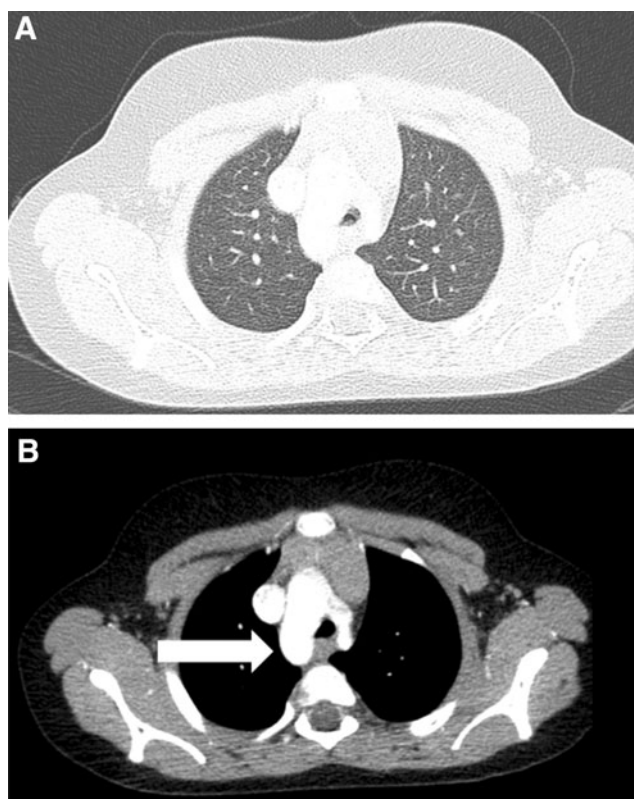


FIG. 3. (A, B) Computed tomography images of the chest with intravenous contrast, with visible right-sided aortic arch (arrow).

with blood flow between the left subclavian artery and the descending aorta, which might correspond to Kommerell's diverticulum. Together with the right-sided aortic arch, this formed a vascular ring around the trachea and esophagus, causing compression and narrowing of the trachea. The arterial ligament was dissected, ligated, and capped, an additional hypoplastic aortic arch was also dissected.

The course of surgery and the postoperative period were uneventful. According to the parental report, previously observed symptoms resolved after the procedure. Exercise tolerance has improved dramatically. Currently, the boy's performance is equal to his peers, and he does not need to make rest breaks during physical activity. The boy remains under cardiologic control.

Informed consent was obtained from the parents of the patient for the publication of his case history.

Discussion

The differential diagnosis of reduced exercise tolerance requires consideration of many potential causes. In case of our patient, spirometry results did not suggest peripheral bronchial obstruction that would be typical for bronchial asthma. On the contrary, the expiratory flow-volume curve was flattened in the proximal segment (the initial stage of expiration). Particular attention was paid to decreased PEF and MEF75 values. These results suggested central airways obstruction. The shape of the inspiratory curve was normal, which preliminarily excluded laryngeal obstruction.^{5,6} No improvement of the expiratory parameters was reported after β_2 -agonist administration, which further reduced the probability of bronchial asthma.

The initial diagnosis was based on a chest X-ray showing a shadow in the right upper mediastinum, probably responsible for the narrowing of the trachea. Chest X-ray still remains the basic examination, justified in the presence of suspected respiratory, cardiovascular, or other chest organ disorders. Ordering this simple test may have contributed to obtaining correct diagnosis before the patient was referred to the hospital. CT unequivocally confirmed the presence of a vascular defect in the form of a right-sided aortic arch modeling the cross-section of the trachea.

The previous cardiologic consultation confirmed parents' belief that their child's discomfort is not related to cardiovascular disorders. However, the interpretation of ECHO depends on many factors, including physician's experience, a thorough topography assessment of vessels arising from the heart. Technical difficulties may affect the course and result of the test, particularly in obese children. Assessment of mediastinal vessels is challenging because of their limited visibility related to bone structures of the chest and lung parenchyma.

Based on the patient's second ECHO report, right-sided aortic arch with the Kommerell's diverticulum was suspected. It was later confirmed intraoperatively. The Kommerell's diverticulum is the widening at the origin of the left subclavian artery arising from the descending aorta or the distal portion of the arch. The Kommerell's diverticulum together with arterial duct (ligament), connecting to the initial segment of the left pulmonary artery, forms a vascular ring

and usually causes compression symptoms (primarily on the trachea, or simultaneously on the trachea and esophagus).^{7,8} A diagram of the anatomical structure of this form of the vascular ring was published by Morel et al. in 2002.⁹

In our patient, complete vascular ring around the trachea and esophagus was associated with compression and narrowing of the trachea. However, the patient did not present with other classical symptoms, suggesting vascular ring, such as stridor, dysphagia, and choking that contributed to the delay in making the correct diagnosis. It was made only during the differential diagnostic process of exercise-related respiratory problems. Spirometry played a crucial role, indicating reduced flows at the central airways level.

In conclusion, various sometimes unexpected diagnoses may simulate bronchial asthma. Spirometry and radiologic chest examination are relevant basic tests in the differential diagnosis workup of reduced exercise tolerance.

Author Disclosure Statement

No competing financial interests exist.

Funding Information

This study is self-funded.

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