Case Report

Demonstration of tracheal stenosis by computed tomography of different modalities and their comparisons: report of one child with congenital stridor and wheezing

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Introduction

Most vascular rings are not associated with intrinsic tracheal anomalies or stenosis other than extrinsic tracheal compression, with the exception of the pulmonary artery sling (1-3). However, we describe an infant presenting with stridor, wheezing and recurrent pneumonia, which is refractory to medical treatment. Preoperative recognition and documentation of intrinsic tracheal stenosis, as well as extrinsic tracheal compression of a vascular ring, which is composed of an aberrant left subclavian artery originating from a right aortic arch, by direct coronal, spiral, and ultrafast or electron beam compute tomography (CT) with three-dimensional reconstruction images, can be therapeutically rewarding in this patient. Respiratory symptoms of wheezing and stridor were alleviated by one-stage cardiothoracic surgery, including division of the aberrant artery and tracheoplaasty with pericardial patch (4).

Case report

A 31-month-old girl, who presented with refractory stridor and wheezing since 6 months of age, came to our medical attention for recurrent pneumonia. She had been operated on for a cleft lip at 3 months old, and for bilateral inguinal hernia and right accessory thumb at 7.5 months of age. On physical examination, she was found dyspnoeic with mouth breathing behavior. Stridor and wheezing were noted in each respiratory cycle. No pathological heart murmur could be heard. Plain chest film showed a right aortic arch shadow.

Oesophagogram revealed a posterior indentation on the oesophagus. Echocardiography did not detect the presence of a double aortic arch. Angiocardiography showed an aberrant left subclavian artery from the right aortic arch. Pulmonary systolic pressure was elevated to 51 mmHg. After meticulous review of direct coronal CT and spiral CT (Fig. 1) from another tertiary medical centre, which failed to inform her parents of a vascular ring composed of an aberrant left subclavian artery from the right aortic arch, and of ultrafast or electron beam CT (Fig. 2), we are very sure that both cardiac and thoracic anomalies can be clearly delineated simultaneously in both modality. One-stage cardiothoracic surgery, including pericardial tracheoplasty, to widen the intrinsic tracheal narrowing, and division of the aberrant artery, to relieve the extrinsic tracheal compression, was performed at 31 months of age. Acute respiratory distress was alleviated remarkably after surgery. In a 2-yr follow-up, she was free of wheezing respiration.

Discussion

With the exception of the pulmonary artery sling, most vascular rings are not associated with intrinsic tracheal anomalies or stenosis other than extrinsic tracheal compression (1-3). Hence it appears that congenital tracheobronchial anomaly and stenosis will not be neglected in patients with pulmonary artery sling for their notorious combinations (2,3). However, in a long-term follow-up of pulmonary functions in postoperative patients with vascular rings and slings, central airway obstruction were found in two patients with a right aortic arch and a left ligamentum arteriosum, in two with anomalous innominate
preoperative recognition and documentation of the tracheobronchial obstruction, due to either intrinsic stenosis or extrinsic compression or both anomalies, in patients with a vascular ring, can make cardiothoracic surgery suitable for all and improve postoperative care. Direct coronal CT (6,7) and spiral volumetric CT (8) have been successfully applied to clearly image the tracheobronchial tree. By the integration of spiral CT techniques and rapid three-dimensional display of tracheobronchial structures and thoracic vascular anatomy, spiral volumetric CT bronchography-angiography becomes practical in evaluating cardiothoracic anomalies simultaneously. The reconstruction images can be obtained without respiratory-induced artifact. Hence rather invasive examinations, such as bronchoscopy, bronchography and angiography, which are not comprehensive in assessing both the thoracic and cardiac anomalies at one time, can be potentially risky and demanding for infants with compromised cardiopulmonary functions. Electron beam CT, or ultrafast CT, has the advantages of high-spatial resolution, non-invasiveness, operator-independence and short scan-
ning time (9). It has been well testified to be a promising complementary modality for an overall understanding of heterotaxy syndrome, in which both intracardiac and extracardiac structures can be clearly delineated at one time (9). We appreciate that direct coronal CT and spiral CT have a more important role in the detection of tracheobronchial airway anomaly rather than the cardiological vessels malformations. We think that the ultrafast or electron beam CT is superior to direct coronal CT and spiral CT in the assessment of both cardiothoracic anomalies in that the high velocity of the cardiac arteries can be overcome in ultrafast or electron beam CT, and then spatial relationship of abnormal anatomy of cardiothoracic vessels and tracheobronchial airway can be much clearly defined simultaneously (9). Ultrafast or electron beam CT can be recommended as a powerful examination modality for a paediatric patients with complex cardiothoracic anomalies, especially in small babies or infants, who are cardiopulmonary distress prone (9).

Intrinsic tracheal stenosis as well as extrinsic tracheal compression must also be considered in an infant with even an aberrant left subclavian artery from a right aortic arch. Preoperative recognition is warranted since one stage cardiothoracic surgery can improve postoperation care. Simultaneous demonstration of cardiac and thoracic anomalies by ultrafast or electron beam CT is feasible.

References


