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Research article

Dual-source computed tomography for quantitative assessment of tracheobronchial anomaly from type IIA pulmonary artery sling in pediatric patients

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ABSTRACT

Purpose: As an extremely rare abnormity, type IIA pulmonary artery sling (PAS) tracheobronchial anomalies should be differentiated from normal airway for therapeutic decision-making. This study aimed to investigate the quantitative characteristics of type IIA PAS tracheobronchial anomalies using dual-source computed tomography (DSCT).

Materials and methods: From January 2009 to December 2016, there were 11 type IIA PAS patients enrolled for analysis and compared with 32 controls. The areas of the trachea, right upper lobar bronchus, left main bronchus, right intermediate bronchus [bridging bronchus (BB) in patients], and subcarinal angle [subpseudocarinal angle in patients] were measured. Measurements other than that for the angle were standardized with body surface area.

Results: Significant differences were found between the type IIA PAS patients and controls in terms of DSCT measurements in the trachea (0.51 ± 0.32 cm^2/m^2 vs. 0.92 ± 0.18 cm^2/m^2, $P < 0.001$); left main bronchus (0.29 ± 0.14 cm^2/m^2 vs. 0.43 ± 0.18 cm^2/m^2, $P = 0.01$); right intermediate bronchus [BB in patients] (0.36 ± 0.13 cm^2/m^2 vs. 0.47 ± 0.12 cm^2/m^2, $P = 0.02$); and subcarinal angle [subpseudocarinal angle in patients] (116.27 ± 16.45° vs. 79.41 ± 15.71°, $P < 0.001$). Receiver operating characteristic analysis further revealed that these parameters may be indicators to differentiate tracheobronchial anomalies due to type IIA PAS from controls (AUC, 0.88–0.98; sensitivity, 81.7%–100%; specificity, 91.7%–100%).

Conclusions: DSCT is an alternative technique to detect tracheobronchial anomalies in pediatric patients with type IIA PAS. It can provide accurate anatomic details for surgeons to determine therapeutic strategies.

1. Introduction

Pulmonary artery sling (PAS) is a rare congenital anomaly that is characterized by an aberrant origin of the left pulmonary artery (LPA) from the posterior aspect of the right pulmonary artery, crossing leftward between the trachea and esophagus to the left pulmonary hilum to form a vascular sling around the trachea [1]. The most probable theory of the embryology of PAS is that there is a caudal capillary connection between the right pulmonary artery and the left post-branchial plexus, which results in an aberrant coursing of the LPA posterior to the tracheobronchial tree [2]. The prevalence of PAS remains controversial because the majority of PAS patients are clinically silent, unless accompanied by other anomalies. Actually, PAS is frequently associated with tracheobronchial anomalies, causing respiratory symptoms, such as wheeze, cough and recurrent respiratory infection, early on in life. However, the underlying embryological association between these two entities has not been clearly reported. In clinical practice, it is the latter mainly determines the outcome of PAS, rather than on the PAS itself [3]. According to Wells' classification of PAS, type IIA lesions refer to the presence of the classic bridging bronchus (BB) [3]. In this type, the tracheobronchial tree mimics the normal airway but the stenosis could endanger pediatric patients. The airway anastomos technique or slide tracheoplasty technique are commonly used to deal with this concern. Therefore, accurate and safe assessment of these anomalies is essential for therapeutic decision-making [4–6].

Cardiac catheterization, transthoracic echocardiography, and

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magnetic resonance angiography are currently used to confirm the diagnosis of PAS. However, these modalities are limited in demonstrating the tracheobronchial tree [2,6,7]. Bronchoscopy is regarded as the gold standard for identifying tracheobronchial abnormalities, but it may result in severe respiratory symptoms for pediatric patients [8]. At present, dual-source computed tomography (DSCT) has been widely utilized in congenital heart disease. With three-dimensional (3D) and other post-processing techniques, it allows visualization of the heart, extravasculature, lungs, and airway. Previous studies reported that multidetector CT was advantageous in delineating the morphologic features of PAS [3,7–9]. However, to the best of our knowledge, the issue on differentiating type IIA PAS tracheobronchial anomalies from normal airway has yet to be reported. In this study, we aimed to investigate the use of DSCT in quantitatively characterizing the tracheobronchial tree in this rare entity.

2. Materials and methods

2.1. Study population

From January 2009 to December 2016, there were approximately 1500 cardiac examinations performed at our medical center. Based on the Wells' classification of PAS, type IIA PAS was diagnosed in 11 patients (6 boys and 5 girls; average age 28.5 ± 31.1 months, range 2–84 months) [3]. For comparison, 32 subjects (18 boys and 14 girls; average age 28.4 ± 28.6 months, range 2–96 months) without respiratory disease were retrospectively selected during the same period. The institutional review board of our hospital approved this study (No. 14–163). Written informed consent, including radiation exposure and adverse reactions to the iodinated contrast agent, was obtained from the guardians of the patients. All patient-sensitive information was treated with full confidentiality and used solely for the purposes of this study.

2.2. Scanning protocol

All subjects were examined using a DSCT scanner (Somatom Definition; Siemens Medical Solutions, Forchheim, Germany). Short-term chloral hydrate (concentration of 10%, 0.5 ml/kg) was given to patients younger than six years of age before examination, whereas older patients were asked to hold their breath during examination. A retrospective electrocardiography-gated protocol was selected for scanning with the following parameters: tube voltage of 80–120 kV (adapted to body mass index), tube current of 220 mAs, gantry rotation time of 0.33 s, and pitch of 0.2–0.5 (adapted to heart rate). Patients received non-ionic contrast agent (Iopamidol 370 mg/ml; Bracco, Italy) at a flow rate of 5 ml/s via an antecubital vein, followed by 20 ml of saline solution at the same flow rate. Acquired data were then processed on a workstation (Syngo; Siemens Medical System, Forchheim, Germany) and the images were reconstructed with a slice thickness of 0.75 mm and an increment of 0.7 mm. Post-processing techniques, such as multiplanar reformation and volume rendering, were used for image analysis (Fig. 1).

2.3. Image analysis

All databases analyses were performed on the Syngo workstation by two experienced radiologists (both were thoracic radiologists with an experience of 5 years) independently. For each PAS patient, the areas of trachea, right upper lobar bronchus, left main bronchus, and BB were measured in order at the maximum site, as well as the subpseudocarinal angle. In a similar way, for each control, the areas of trachea, right upper lobar bronchus, left main bronchus, and the right intermediate bronchus were measured in order at the maximum site, as well as the subcarinal angle. The BB and the subpseudocarinal angle (the angle between the BB and left main bronchus)in the study patients were considered to correspond with the right intermediate bronchus (branching out into right middle and inferior lobar bronchus) and the subcarinal angle (the angle between right main bronchus and left main bronchus) in controls, respectively (Fig. 2). All the measurements for each individual were accomplished within 30 min and values other than that for the angle were standardized by body surface area (BSA). The associated cardiac malformations demonstrated by DSCT were also recorded.

To determine intraobserver variability of DSCT measurements, one radiologist initially measured these parameters and repeated the same work a month later. To determine interobserver variability of DSCT measurements, another radiologist who was blinded to the results reanalyzed the data.

2.4. Radiation dose estimation

The volume CT dose index and dose-length product were automatically displayed on the CT console after examination. The effective dose (ED) was calculated based on the size-specific dose estimates (SSDEs) [10–12].

2.5. Statistical analysis

Acquired data was analyzed by using SPSS software (version 17.0 for Windows, SPSS, Chicago, IL, USA) and MedCalc software (version 9.3.0.0, MedCalc software, Mariakerke, Belgium). Continuous data were presented as mean ± standard deviation, whereas categorical data were presented as numbers and percentages. Student's *t*-test and Chi-Square test were used as appropriate to assess the differences in baseline characteristics and quantitative parameters between patients and controls. Receiver operating characteristic (ROC) was used to further predict the sensitivity and specificity of those parameters in differentiating type IIA PAS patients with tracheobronchial abnormalities from controls. Intraclass correlation coefficients (ICCs) were calculated to assess the intra- and inter-observer variability. P value of < 0.05 was considered statistically significant.

3. Results

3.1. Baseline characteristics

The basic characteristics of 11 patients with type IIA PAS and 32 controls are listed in Table 1. There were no significant differences in age, gender, height, weight, heart rate, systolic, and diastolic blood pressure between the two groups (all $P > 0.05$). Among the PAS patients, there were 6 cases (54.5%) with fever, 6 cases (54.5%) with wheeze, 4 cases (36.4%) with cough, and 3 cases (27.3%) with dyspnea upon referral to our medical center. Additionally, 4 cases received LPA reimplantation operation; 1 case received both LPA reimplantation operation and airway resection combined with anastomosis technique.

3.2. Quantitative analysis of tracheobronchial structures

The measured parameters between the two groups were indexed by BSA and then summarized in Table 2. Significant differences in the areas of the trachea ($0.51 \pm 0.32\ cm^2/m^2$ vs. $0.92 \pm 0.18\ cm^2/m^2$, $P < 0.001$); left main bronchus ($0.29 \pm 0.14\ cm^2/m^2$ vs. $0.43 \pm 0.18\ cm^2/m^2$, $P = 0.01$); right intermediate bronchus [BB in PAS patients] ($0.36 \pm 0.13\ cm^2/m^2$ vs. $0.47 \pm 0.12\ cm^2/m^2$, $P = 0.02$); and subcarinal angle [subpseudocarinal angle in PAS patients] ($116.27 \pm 16.45^\circ$ vs. $79.41 \pm 15.71^\circ$, $P < 0.001$) were found between PAS patients and controls. However, statistical analysis showed no significant difference between PAS patients and controls in the area of the right upper lobar bronchus ($0.28 \pm 0.15\ cm^2/m^2$ vs. $0.28 \pm 0.10\ cm^2/m^2$, $P = 0.89$).