A Case of Combined Parathracheal Air Cyst and Accessory Cardiac Bronchus

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Paratracheal air cyst (PTAC) is rather frequently detected on thoracic multi-detector computed tomography (MDCT) in daily practice. Accessory cardiac bronchus (ACB) is a rare anomaly; however, the incidence rate is increasing with the use of recent high quality MDCT scanners. We report a case of combined PTAC and ACB that was incidentally detected by MDCT. Three dimensional CT images revealed anatomical details.

Key words: paratracheal air cyst, accessory cardiac bronchus, MDCT

INTRODUCTION

PTAC is a small air collection in the right paratracheal area at the level of the thoracic inlet. PTACs comprise various pathological entities [1, 2]. Majority of PTACs reported in the literature are diagnosed on CT as tracheal diverticula with PTACs connected to the trachea via a communicating channel [3, 4]. CT examinations reveal an incidence of 2%–8.1% [1, 2, 5, 6]. ACB is a congenital bronchial anomaly with an incidence of 0.08%–0.57% [7, 8]. Both anomalies are usually asymptomatic and detected incidentally on CT imaging. We report a rare case of combined PTAC and ACB in a female patient without a respiratory disorder.

CASE REPORT

A 43-year-old Japanese female underwent a CT scan using MDCT to rule out lung metastasis from carcinoma of uterine cervix. She had no clinical symptoms. She was uneventful during pregnancy and delivery. She had no history of respiratory tract infection, asthma and tracheal intubation. We used a 128 MDCT scanner (SOMATOM Definition Flash: SIEMENS Forchheim, Germany) with a slice thickness of 0.5 mm. The axial CT image at the level of thoracic inlet showed a small lobulated air cyst measuring 5 mm in the right paratracheal region (Figure a). The axial CT image at a level inferior to the bifurcation of the trachea showed an anomalous bronchus originating from the intermediate bronchus (Figure b). The lung around the anomalous bronchus was clear. A virtual bronchoscopic image showed communication between the air cyst and the trachea (Figure c). Three dimensional CT images including a volume rendering image and a virtual bronchoscopic image clearly revealed both anomalies (Figure b–d). The volume rendering image showed that an anomalous bronchus had a blind extremity (Figure d). The CT findings were compatible with PTCA and ACB.

DISCUSSION

PTACs are collections of air adjacent to the trachea that are usually silent and detected incidentally. The histopathological diagnosis of PTACs in reported, surgically confirmed cases include tracheal diverticulum, trachecele, lymphoepithelial cyst, and bronchogenic cyst [2]. Most studies have described the majority of PTACs as tracheal diverticula due to their connection with the trachea [2]. In this case, the PTAC is a tracheal diverticulum, because the connection between the cyst with the trachea could be shown on MDCT. Tracheal diverticula and bronchial diverticula are similarly generated from mucosal herniation through a weakened tracheobronchial wall [9]. Tracheal diverticula are divided into congenital and acquired forms. Although the former is considered to develop from a developmental defect in the tracheal cartilage, the latter is considered to be due to increased intraluminal pressure caused by chronic obstructive pulmonary disease (COPD) [1, 5]. Congenital tracheal diverticula, comprising respiratory epithelia, smooth muscle, and cartilage are smaller and have a narrow communication with the trachea [10, 11]. Congenital tracheal diverticula are thought to represent a vestigial supernumerary lung or an aborted, abnormally high division of the primary lung bud [11]. Acquired tracheal diverticula are larger with a wide opening and comprise respiratory epithelia only [10]. PTACs are primarily located at the level of the thoracic inlet along the right posterolateral trachea [1, 2, 10]. Congenital tracheal diverticula appear 4 cm–5 cm below the vocal cords or a few centimeters above the carina [11]. Acquired tracheal diverticula may appear at any level [10, 11]. Therefore, this case may be a congenital tracheal diverticulum because the cyst size was small with a narrow tracheal connection and the patient had no cause of increased airway pressure such as COPD or chronic cough; however, pathological confirmation was not possible. Bae *et al.* reported that

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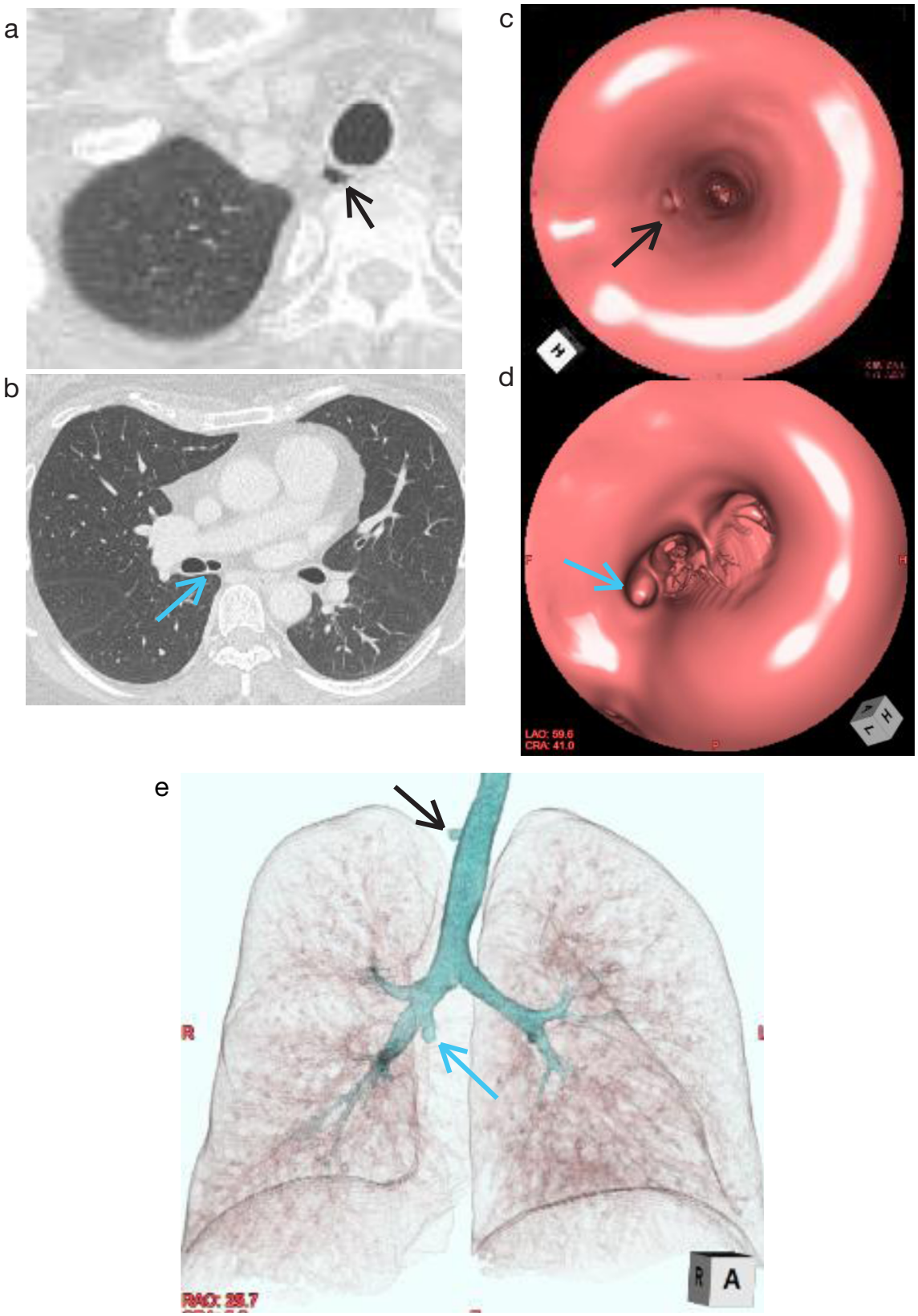

Figure a. The axial CT image at the level of thoracic inlet shows a small lobulated air cyst in the right posterolateral tracheal region (black arrow). b. The axial CT image at the level inferior to the bifurcation of the trachea shows abnormal air on the left side of the intermediate bronchus (blue arrow) surrounded by a normal lung. c. Virtual bronchoscopic image of the trachea demonstrates communication between the air cyst and the trachea. d. Virtual bronchoscopic image of the right bronchus shows a small air pouch (blue arrow). e. Volume rendering image reveals both air components (black and blue arrows) clearly and an anomalous bronchus of blind extremity (Figure d).

the size of PTACs range from 1.5 mm to 24 mm on the axial CT image and 37.7% of PTACs show a connection with the trachea using a 3 mm–5 mm imaging slice thickness [2]. The incidence of PTAC was 2%–8.1% on MDCT. The difference among the studies is due to the thinner cuts of scans and higher resolution [1, 2, 5, 6]. Some authors studied the PTAC association with COPD [1, 2, 5, 6]. While some results concluded that PTACs are not correlated with the presence of COPD [2, 6], the largest study by Polat *et al.*, in 2014 showed that the relation between PTACs and COPD was statistically significant [1].

Major bronchial abnormalities include ACB and tracheal bronchus [7]. ACB is the only true supernumerary anomalous bronchus. It conically progresses from 1 cm to 5 cm in a caudal direction toward the pericardium, paralleling the intermediate bronchus [7]. The length of the ACB is variable, ranging from a short, blind-ending diverticulum to a longer branching structure [12]. The short type is usually a simple bronchial stump without associated alveolar tissue such as in this case, whereas the longer subtype has been reported with and without associated rudimentary alveolar tissue [12]. Some cases had ventilated lobules or anomalous fissures [7]. ACB is a developmental aberration of bronchial branching, likely occurring between the 4th and 6th weeks of embryonic life [13]. Shizuki *et al.* reported associations with the absence of medial basilar bronchus [8]. Some reports include simultaneous occurrence of ACB and tracheal bronchi [14, 15]. Majority of ACBs is asymptomatic, rarely this blind-ending airway serve as a potential reservoir for infectious material. In these cases, clinical symptoms are thought to relate to retained secretions with resultant inflammation, hypervascularity, and hemoptysis [12].

To the best of our knowledge, there is no reported case of combined PTAC and ACB. We suspect that the association with both congenital tracheobronchial abnormalities is incidental. However, a limitation of the present case was that pathological confirmation was impossible. Both congenital abnormalities are commonly discovered as an asymptomatic finding, and a MDCT is frequently used for the detection of those abnormalities. An important differential diagnosis of congenital PTAC is pneumomediastinum [1]. Differential diagnoses of ACB include acquired bronchoesophageal fistula and traction bronchiectasis due to lymphadenopathy [8]. The best modality for diagnosis of congenital minor anomalies is high quality CT images by MDCT, such as mutiplanar reconstruction and 3D CT images, including virtual bronchoscopy and volume rendered images. Thereafter, exact diagnosis can be achieved.

CONCLUSION

We reported a rare case of combined PTAC and ACB that was incidentally detected using 128 MDCT. Three dimensional CT and thinner 2D images were able to reveal the anatomy clearly and useful for the diagnosis.

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