Role of multi slice computed tomography in the evaluation of congenital anomalies of tracheobronchial tree and lungs

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Abstract  Objective: To evaluate the role of MSCT in the evaluation of congenital anomalies of tracheobronchial tree and lung

Patients and methods: Twenty nine patients with congenital anomalies of tracheobronchial tree and lung were examined using four and six MDCT.

Results: Seven patients (24%) had congenital cystic adenomatoid malformation where successful differentiation of the type was possible. Three patients (10.3%) with bronchopulmonary sequestration were categorized as two (7%) intralobar and one (3.5%) extralobar sequestrations. Four patients (14%) had congenital lobar overinflation. Four patients had Scimitar syndrome with right pulmonary artery hypoplasia, right sided lung hypoplasia, anomalous arterial supply and anomalous pulmonary venous drainage together with pulmonary hypertension. Abnormal bronchial anatomy was revealed in two patients with pulmonary isomerism and one patient with situs inversus. Bronchogenic cyst, tracheal bronchus, Kertagner’s syndrome, pulmonary agenesis and horse shoe lung were seen in one patient each. Dynamic MSCT shows the lunate configuration of the trachea during forced expiration in one patient with tracheomalacia. In one patient with tracheo-esophageal...
1. Introduction

Congenital anomalies of the chest are an important cause of morbidity in infants, children and even adults (1). Classification of the congenital anomalies of the tracheobronchopulmonary apparatus is controversial (2). Common developmental lung anomalies are classified into three broad categories. Bronchopulmonary (lung bud) anomalies including pulmonary agenesis, congenital bronchial atresia, congenital lobar emphysema, congenital cystic adenomatoid malformation (CCAM), pulmonary bronchogenic cysts, tracheal bronchus, tracheomalacia, accessory cardiac bronchus, pulmonary isomerism, Kartagener’s syndrome, cystic fibrosis and tracheo-esophageal fistula. Vascular anomalies including interruption or absence of a main pulmonary artery, anomalous origin of the left pulmonary artery from the right, partial or complete anomalous pulmonary venous drainage and pulmonary arteriovenous malformation. Combined lung and vascular anomalies including hypogenetic lung (Scimitar) syndrome with or without horse shoe lung and bronchopulmonary sequestration, both intralobar and extralobar (3).

MDCT has broadened the imaging of lung anatomy down to the subsegmental level by offering various reformation techniques, including multiplanar reconstruction, shaded-surface display (SSD), minimum-intensity projection, maximum-intensity projection, sliding thin slab imaging, volume rendering and virtual bronchoscopy (4). The aim of this work was to evaluate the role of MSCT in congenital anomalies of tracheobronchial tree and lungs.

2. Patients and methods

This study included 29 patients with congenital anomalies of the lungs or tracheobronchial tree. All patients had multidetector row CT of the chest after IV contrast injection except one patient with, moderate renal insufficiency, scanned without contrast injection. Patients with mild renal insufficiency were aggressively prepared. Examinations were done on GE Light speed 4 scanner; GE Medical Systems, Milwaukee, WI, USA and Siemens Emotion 6 MDCT; Siemens, Erlangen, Germany. Scanning parameters were: volumetric high-spatial-frequency kernel algorithm. Slice thickness: 1.25 mm. Table speed for volumetric HRCT to enable the least cycles of breath-holds as possible. Tube rotation: 0.6–0.9 s (0.75 s); detector collimation 1 mm; helical mode (volumetric HRCT) and field of view (FOV) for small, medium and large patients. kVp and mA per slice: 80–100 kVp and approximately 200–240 mA, although lower doses (60–90 mAs) were used with pediatric and small patients or those receiving serial HRCT scans. For adequate multiplanar reconstruction; scans were performed to cover the root of the neck down to the level of the adrenal glands. The thin slices were sent to the workstation to be viewed in the axial, sagittal and coronal planes as well as for displaying volume rendering. The imaging studies were reviewed by two radiologists.

3. Results

Twenty nine patients, 15 males and 14 females, with congenital bronchopulmonary anomalies were studied. Their ages ranged between 1 day and 14 years with a mean age of 6 months to 1 year. Respiratory distress, tachypnea, wheezes and chest retractions were the common presentations; however, failure to thrive and feeding difficulty were also encountered.

The study included seven patients with congenital cystic adenomatoid malformation, four patients with congenital lobar emphysema, four patients with Scimitar syndrome, three patients with bronchopulmonary sequestration and three patients with pulmonary isomerism. The rest of the patients were distributed through the other diagnoses as shown in Table 1.

Congenital bronchopulmonary anomalies were incidentally discovered in four patients performing CT chest, whereas the rest of the patients were recommended for CT after being examined by a chest X-ray to exclude congenital bronchopulmonary anomalies; of the four patients, two patients (7%) had pulmonary isomerism of the bilateral left lung type associated with polysplenia, one patient (3.5%) had complete situs inversus and one patient (3.5%) had pulmonary bronchogenic cyst.

The seven patients (24%) with congenital cystic adenomatoid malformation were subdivided into five patients (17.2%) with congenital cystic adenomatoid malformation type I, one (3.5%) with congenital cystic adenomatoid malformation type II and one (3.5%) with congenital cystic adenomatoid malformation type III. CT findings of congenital cystic adenomatoid malformation type I included single or multiple air filled cystic lesions larger than 2 cm in diameter and surrounded by small cysts (Fig. 2). Congenital cystic adenomatoid malformation type II appears as air or fluid filled multicystic lesion less than 2 cm in diameter (Fig. 3), while congenital cystic adenomatoid malformation type III appeared a solid lesion.

Four patients with congenital lobar emphysema were included; of them two showed left lung affection and two showed right lung affection. CT revealed overinflation and increased lucency of a lobe or a segment of the lung associated with stretching and attenuation of the parenchymal pulmonary vasculature and mediastinal shift to the contralateral side.

Three patients (10.5%) had bronchopulmonary sequestration; of which two (7%) were intralobar and one (3.5%) was extralobar. On CT, the intralobar type appeared as a bizarre increased lucency or a mass lesion involving the left lower lobe with anomalous arterial blood supply from abdominal aorta and venous drainage into the inferior pulmonary vein. The extralobar type appeared as a left lower lobe lesion with anomalous arterial blood supply from abdominal aorta and venous drainage into azygous vein (Fig. 4).

Four patients (13.7%) had congenital venolobar pulmonary or Scimitar syndrome. CT showed right pulmonary artery...
hypoplasia, right sided lung hypoplasia, anomalous arterial blood supply from the abdominal aorta without sequestration and anomalous pulmonary venous drainage into infradiaphragmatic IVC (Scimitar vein) (Fig. 6).

One hybrid lesion (3.5%) was encountered consisting of congenital cystic adenomatoid malformation type II and intralobar sequestration. The sequestrated lobe appeared on CT as a moderately enhancing lesion involving the left posterior basal segment with anomalous arterial blood supply from the aorta and venous drainage into inferior pulmonary vein. The congenital cystic adenomatoid malformation type II appeared as a multilocular air filled cystic lesion involving most of the left lower lobe and was intimately related to the sequestrated lobe (Fig. 5).

Horse shoe lung was seen in one patient (3.5%), CT showed a portion of the right lower lobe crossing the midline; posterior to the heart and anterior to the esophagus and descending aorta and apparently fused with the left lower lobe.

Only one patient (3.5%) with bronchogenic cyst was encountered accidentally in a patient with renal insufficiency performing CT chest for follow up on pulmonary edema, and appeared as a well-defined unilocular fluid filled cystic lesion involving the right lower lobe.

Kertagnier’s syndrome was seen in one patient (3.5%), and was revealed as left upper and lower lobes bronchiectasis as well as inversion of the bronchial anatomy with right sided hyparterial bronchus, i.e., below right main bronchial artery and left sided eparterial bronchus, i.e., above the left main pulmonary artery. This was associated with dextrocardia and right sided aortic arch. Abdominal scans showed complete situs inversus.

Pulmonary isomerism was encountered in two patients (7%) and complete situs inversus was encountered in one patient (3.5%). The three cases were asymptomatic as regards the abnormal bronchial anatomy and were incidentally discovered. Pulmonary isomerism was encountered in two patients with polyspleenia and situs ambiguous where CT showed the left and right main bronchi passing below the main pulmonary arteries (hyparterial bronchus), i.e., the main stem bronchi as well as their segmental and subsegmental divisions and the lung lobes are of the left lung type. Abnormal bronchial division was also encountered as a part of complete situs inversus diagnosed in one of the three patients. CT showed the right main bronchus passing below the right main pulmonary artery, i.e., hyparterial bronchus while the left main bronchus passing above the left main pulmonary artery, i.e., eparterial bronchus and consequently the lung lobar divisions are inverted with the right lung consisting of two lobes and the left lung consisting of three lobes which is totally opposite to the anatomy (Fig. 1).

In the only one patient (3.5%) with pulmonary agenesis, CT revealed complete absence of the right lung with rudimentary right main bronchus consistent with group 2 of pulmonary underdevelopment (pulmonary aplasia) according to Schneider and Schwalle classifications. This was associated with marked mediastinal shift to the right side together with compensatory hypertrophy of the left lung (Fig. 8).

Tracheal bronchus was encountered in one patient (3.5%) where CT showed prearterial (true right tracheal bronchus) arising from the trachea just above the carina and ventilating mostly the apical segment of the right upper lobe.

One patient (3.5%) with tracheomalacia was presented where CT revealed complete inflation of the trachea at the end of forced inspiration while at the end of forced expiration complete flattening mounting to near total collapse with lunate configuration of the trachea was noted (Fig. 7).

Tracheo-esophageal fistula was seen in a one month old infant (3.5%) with history of near total esophagectomy for tracheo-esophageal fistula leaving the lower third of the esophagus and gastrostomy for current feeding and future reconstruction by colonic bypass but the infant still suffered from aspiration pneumonia. CT following contrast injection into gastrostomy tube showed a fistulous tract connecting the lower third of the esophagus (left during operation) with the carina with opacification of the right main bronchus and its branches. There were also consolidative patches involving both right upper and lower lobes denoting aspiration pneumonia.

4. Discussion

Congenital lung anomalies vary in their clinical manifestations and imaging appearance. Although radiographs play a role in the initial imaging evaluation of patients with clinical suspicion of congenital lung anomalies, cross-sectional imaging is required for the confirmation of diagnosis, further characterization and preoperative evaluation in surgical lesions (5).

The combination of fast speed, high spatial resolution, and enhanced quality of the multi-planar reformation and three-dimensional reconstructions such as CT bronchography and virtual bronchoscopy makes multi-detector CT ideal for evaluating congenital lung anomalies (5).

The present study included 29 patients with congenital bronchopulmonary anomalies. Examinations were performed on MSCT scanners and the acquired images were sent to a picture archiving and communication system “PACS system”; GE Healthcare Technologies for viewing on highly specialized workstations. Multi-planar reconstruction of the acquired thin sliced axial images facilitated not only coronal and sagittal viewing but also 360° oblique views in certain circumstances to trace a bronchus in question. MinIP rendered views; CT bronchography and virtual bronchoscopy were also practical in better appreciation of the airway anomalies.

Congenital cystic adenomatoid malformation accounts for 25% of all congenital lung abnormalities and consists of

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<th>Final diagnosis</th>
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<td>3.5</td>
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<tr>
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adenomatoid proliferation of bronchioles that form cysts instead of normal alveoli. Three types have been classified by pathologic analysis. Type I consists of cysts measuring 2–10 cm. Type II has numerous smaller, more uniform cysts measuring 0.5–2 cm in diameter, and type III are solid-appearing lesions that microscopically demonstrate tiny cysts (3). Type I congenital cystic adenomatoid malformations are the most common type, constituting of approximately 70% of cases. Type II congenital cystic adenomatoid malformation makes up around 15–20% of cases. Type III CCAMs are rarely seen postnatally and have poor prognosis (6). An internal air-fluid level and enhancing thick wall can be seen in infected CCAM

Figure 1  A 1 year old female patient with pulmonary isomerism, polysplenia and dextrocardia. (a and b) Axial CT images showing the left and right main bronchi passing below the main pulmonary arteries (hyparterial bronchus) as shown by the red arrows. (c) VR reconstructed CT image showing associated patent ductus arteriosus (PDA) as shown by the yellow arrow. (d) Sagittal reformatted CT image showing coarctation of the aortic arch in the same patient (orange arrow). (e) Axial CT image showing dextrocardia with the apex of the heart directed to the right side. (f) Axial CT scan through the abdomen shows multiple spleunules noted on the left side (yellow arrows).
Older infants and children present with recurrent infections of CCAM which must be differentiated from necrotizing pneumonia (6).

Congenital lobar overinflation accounts for 10–15% of congenital masses of the lung with more than 90% of infants with CLO presenting with progressive respiratory distress with typical presentation within the 1st 6 months of life (8). CLO occurs secondary to a ball-valve mechanism (9). Bronchomalacia caused by a deficiency of bronchial cartilage, bronchostenosis, bronchotorision, obstructive mucosal flaps or mucosal thickening, cartilaginous septa and bronchial atresia have all been described pathologically in congenital lobar overinflation lobectomy specimens. Some authors prefer the expression congenital lobar overinflation to congenital lobar emphysema; because microscopically, congenital lobar overinflation specimens do not always demonstrate alveolar destruction but rather overdistended but intact alveoli (10,11).

The most common components of congenital pulmonary venolobar or Scimitar syndrome are hypogenetic lung and anomalous pulmonary venous return (12). Isolated anomalous systemic supply to the lower lobes without bronchial abnormality is also reported (13). In patients in whom hypoplastic lung tissue is present, the proximal portion of the ipsilateral pulmonary artery is absent; however, the peripheral pulmonary arteries are present. Systemic collateral circulation to the peripheral pulmonary arteries occurs and most often arises from the bronchial arteries. With time, large transpleural collateral vessels to the intercostal arteries may also develop (14). The anomalous pulmonary vein most often drains into the IVC below the right hemidiaphragm. Less commonly PAPVR drains into the suprahepatic portion of the IVC, hepatic veins, portal vein, azygous vein, coronary sinus or right atrium. Drainage of the anomalous vein into the suprahepatic portion of the IVC or right atrium may be a clue to associated congenital interruption of the intrahepatic portion of the IVC. When PAPVR drains to a systemic vein or to the right atrium, there is a left-to-right shunt, which is usually asymptomatic unless the shunt is 2:1 or greater. On occasion, the PAPVR drains to the left atrium which when occurs; the anomalous vein is called a “meandering” pulmonary vein (15).

Three patients with bronchopulmonary sequestration were included. The intralobar sequestration accounts for 75% while extralobar sequestration accounts for 25% of all the cases of bronchopulmonary sequestration (5). Intralobar sequestration

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**Figure 2**  A 7 months old female patient with congenital cystic adenomatoid malformation type I. (a) axial CT image shows a unilocular air filled cystic lesion involving the right lower lobe averaging about 5 cm in diameter. (b) Coronal reformatted CT image showing the cystic lesion involving the right lower lobe.

**Figure 3**  A 6 months old female patient with congenital cystic adenomatoid malformation type II. (a) Axial CT images show a multilocular fluid filled cystic lesion involving most of the right lower lung lobe. (b) Axial CT images with lung window/level show associated air fill multilocular cystic component as a part of the whole cystic mass lesion. Mediastinal shift to the left side is also noted.
may appear as a homogenous well defined mass lesion or multicycstic lesion, either as an air or fluid filled lesion or hyperlucent and hypovascular region of the lung while the extralobar sequestration appears commonly as a well defined homogenous mass lesion that rarely contains air (16). The systemic supply to the lesion is commonly from a separate branch from the aorta, but upper abdominal vessels and even coronary arteries are also described (17). Extralobar sequestration is commonly associated with other congenital malformations in over 50% of cases, most noticeably congenital diaphragmatic hernias; congenital heart disease; and CCAM type II (hybrid lesions) (18). The embryology of both ILPS and ELPS is disputed (19). Most authors agree that ELPS is a true developmental anomaly that arises from a supernumerary lung bud from the primitive foregut. This connection may persist, giving rise to a communication with the gastrointestinal tract. Some authors argue that ILPS are acquired lesions that arise in chronically infected lung segments, with bronchial occlusion and a parasitized systemic arterial supply to support a compromised segment of lung.

One hybrid lesion was encountered consisting of CCAM type II and intralobar sequestration. Pulmonary sequestration may coexist with congenital cystic adenomatoid malformation. Extralobar pulmonary sequestration is reported to exist with type II CCAM in up to 50% of cases. Pulmonary sequestration and congenital cystic adenomatoid malformation likely
represent part of a spectrum of related congenital lung anomalies. The radiologist needs to search for an aberrant systemic arterial vessel when imaging suspected cases of CCAM (20,21).

Horse shoe lung is an uncommon malformation that is typically associated with the hypogenetic lung syndrome and all variants of the lobar agenesis–aplasia complex in which a portion of the right lower lobe crosses the midline and is fused with the left lower lobe. This isthmus of pulmonary tissue is posterior to the heart and anterior to the esophagus and descending aorta. The right and left lower lobes may fuse, either with or without intervening layers of visceral pleura. Typically they are sheathed in a continuous layer of parietal pleura, forming a communication between the right and left pleural cavities. In most cases, the arterial supply to the isthmus is from an anomalous branch of the right pulmonary artery. The bronchi arise from the right bronchial tree (21,22).

Bronchogenic cyst was encountered accidentally in a patient with renal insufficiency performing CT chest for follow up on pulmonary edema. Bronchogenic cysts of the lung originate from the foregut, as do such cysts in the mediastinum. The foregut cysts develop within the cleavage between the respiratory tract and the digestive tube. When they form early, they are located in the mediastinum by the trachea and esophagus or close to the carina and main bronchi. When they occur

Figure 5  A 3 months old male patient with hybrid lesion consisting of intralobar sequestration and congenital cystic adenomatoid malformation type II. (a–c) Contrast enhanced axial CT images showing intralobar sequestrated lobe (blue arrows) as moderately enhancing lesion involving the posterior basal segment of the left lower lobe with anomalous arterial blood supply from the aorta (red arrows) and venous drainage into inferior pulmonary vein (green arrows). (d) Axial CT image with lung window/level showing associated congenital cystic adenomatoid malformation type II as a multilocular air filled cystic lesion involving most of the left lower lobe and related to the sequestrated lobe. (e) VR reconstructed CT image showing the sequestrated lobe with its anomalous arterial blood supply and its venous drainage.
later, during bronchial budding and branching, they grow within the lung parenchyma (17). Most bronchogenic cysts can be confidently diagnosed by using non-enhanced CT; however, administration of contrast material at CT or the addition of MRI can be useful for differentiating problematic soft-tissue-attenuation cysts with internal heterogeneity, high attenuation numbers, streak artifact or atypical location from mediastinal neoplasia. Bronchogenic cysts are more common in the lower lung lobes; in particular, the subpleural region of the lower lobe (23).

Kertagener’s syndrome is the most dramatic manifestation of primary ciliary dyskinesia (PCD). It presents with complete thoracic and abdominal situs inversus, lower lobe predominant bronchiectasis (most often) and sinusitis. Chronic middle ear infections and infertility are frequently associations. The normal ciliary function is required during intrauterine fetal development for normal foregut rotation and the development of normal thoracic and abdominal situs. For this reason, half of patients with PCD have been believed to have complete situs inversus and half to have normal situs. PCD patients with
normal situs can share all the same maladies of Kertagener’s syndrome minus the situs abnormality. However, recently, it has been noted that several of the ciliary deficiencies may not cause total immotility and all ultrastructural variants are not associated with situs inversus in 50% of cases. Kertagener’s syndrome usually presents with lower lobe predominant bronchiectasis. The bronchiectasis detected by MDCT in Kertagener’s syndrome is more likely to be cylindrical than varicoid or cystic. There is no preferential involvement of the central or peripheral airways with bronchiectasis (24,25). MSCT evaluation of bronchiectasis is appreciably replacing the gold standard of conventional HRCT in view of detection and confidence of diagnosis of small size bronchiectatic changes (26–28).

Pulmonary isomerism was encountered in two patients and complete situs inversus in one patient. Pulmonary isomerism was encountered in two patients with polysplenia and situs ambiguous. Abnormal bronchial division was also encountered as a part of complete situs inversus diagnosed in one of the three patients. Pulmonary isomerism is an anomaly of the number of lung lobes. In the common variety of pulmonary isomerism, the right lung has two lobes, whereas the left has three lobes. This anomaly may be associated with situs inversus, asplenia, polysplenia and/or anomalous pulmonary drainage (29).

Only one case of pulmonary agenesis was encountered. Pulmonary underdevelopment is classified into three groups: group 1, bronchus and lung are absent (agenesis); group 2, a rudimentary bronchus is present and limited to a blind-end pouch without lung tissue (aplasia) and group 3, there is bronchial hypoplasia with variable reduction of lung tissue (hypoplasia) (30). Cunningham et al. (31) presented eight cases, all of which had associated congenital abnormalities of other systems on the same side as the agenesis. They hypothesize an abnormality of blood flow in the dorsal aortic arch during the 4th week of gestation to be the cause. Unilateral lung agenesis–aplasia occurs with equal frequency on both the right and left sides. Bilateral agenesis–aplasia is obviously fatal (32).

Right-sided agenesis is usually more symptomatic even in the absence of associated anomalies, due to mechanical compression of the left main bronchus with possible bronchomalacia (33). The major cardiovascular anomalies have included atrial/ventricular septal defects, patent ductus arteriosus, tetralogy of Fallot, single ventricle, partial or total anomalous pulmonary venous return, persistent left superior vena cava and inferior vena cava drainage into the left atrium, which is more common in patients with absent right lung.

The term tracheal bronchus includes a variety of bronchial anomalies arising from the trachea or main bronchus and directed toward the upper-lobe territory. The anomalous bronchus usually exits the right lateral wall of the trachea less than 2 cm above the major carina and can supply the entire upper lobe or its apical segment (4). The tracheal bronchus...
may be displaced or supernumerary (34). If the anatomic upper-lobe bronchus is missing a single branch, the tracheal bronchus is defined as displaced; if the right upper-lobe bronchus has a normal trifurcation into apical, posterior, and anterior segmental bronchi, the tracheal bronchus is defined as supernumerary. The supernumerary bronchi may end blindly; in that case, they are also called tracheal diverticula. If they end in aerated or bronchiectatic lung tissue, they are termed apical accessory lungs or tracheal lobes. Most of these bronchial branching anomalies are well diagnosed at chest CT as a linear area of hypoattenuation arising directly from the trachea. The displaced type of tracheal bronchus is more frequent than the supernumerary type. This fact may be well demonstrated with high-resolution spiral CT, which can demonstrate that the aberrant bronchus may correspond to a segmental, subsegmental or subsubsegmental bronchus (35).

Tracheomalacia is a condition of the neonatal and infant airway characterized by weakness of the supporting tracheal cartilage and widening of the posterior membranous wall; all causing tracheal collapse especially during increased airflow, such as coughing, crying or feeding. Tracheomalacia most commonly affects the distal one-third of the trachea (36).

Tracheo-esophageal fistula was seen in one patient. Esophageal atresia with or without tracheo-esophageal fistula represent a complex of congenital anomalies characterized by failure of formation of the tubular esophagus and/or an abnormal communication between the esophagus and trachea. It is thought to be due to a developmental disorder in formation and separation of the primitive foregut into trachea and esophagus (37,38).

Our experience in assessing the role of high-resolution and MDCT may have been limited by the non-availability of several other congenital bronchopulmonary anomalies, however, we believe that MSCT has proved to be indispensable in assessment, staging and preoperative planning of different types of congenital bronchopulmonary anomalies.

References