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HYBRID CHEST LESION IN A NEONATE: IMAGING RADIOLOGICAL FEATURES ON CT ANGIOGRAPHY.

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ABSTRACT

Hybrid chest lesion is an uncommon congenital thoracic anomaly. We report a case of hybrid chest lesion in a neonate, who was initially treated for congenital pneumonia. The initial computed tomography thorax in portovenous phase was performed due to persistent opacities at right mid and lower zones on chest radiograph. The echocardiographic findings were suspicious of congenital cardiovascular anomalies. The subsequent ECG-gated CT angiography thorax demonstrated features consistent with hybrid lesion, together with its vascular anomalies which were confirmed intraoperatively and histologically. The baby underwent a successful surgery to resect the sequestrated lobe with plication of the right diaphragm and is doing well.

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Keywords: Bronchopulmonary Sequestration, Hybrid Chest Lesion; Computed Tomography Angiography; Congenital Anomalies, Thoracic; Neonate; Pneumonia, Congenital.

INTRODUCTION

Extralobar sequestration associated with congenital pulmonary airway malformation, known as hybrid lesion is an extremely rare congenital pulmonary disorder.[1] This condition can be diagnosed antenatally in utero by prenatal ultrasound or magnetic resonance imaging (MRI).[2,3] Postnatally, diagnosis is usually made following investigations for recurrent complications such as pneumonia, lung abscesses or pneumothorax. Computed tomography angiography (CTA) of thorax may be needed for further characterisation and better delineation of vascular structures, which is important for surgical planning.[4,5] In this case, we aim to discuss the imaging features of a hybrid chest lesion and the benefit of postnatal ECG-gated CTA thorax prior to surgical intervention.

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CASE REPORT

This is a case of a full term baby girl who was born well via spontaneous vaginal delivery. Antenatally, besides maternal iron deficiency anaemia and gestational hypertension, the fetus has been well. The baby was referred to the paediatric team at 2 hours of life for respiratory distress syndrome. She was initially treated for congenital pneumonia, requiring

ventilation support. However, her condition deteriorated despite completing one cycle of intravenous antibiotic. Serial chest radiographs showed persistent right mid to lower zone opacities (Figure 1). Several differential diagnoses were considered at this stage and listed in Table I.

Initial contrast-enhanced computed tomography (CT) thorax at the primary hospital showed enhanced soft tissue mass in the right lower lobe associated with tenting of the hemidiaphragm but no significant mediastinal shift. The venous drainage of this mass was directed into the main portal vein with no obvious arterial supply seen (scan was only done in plain and portal venous phases). This led to the conclusion of a right extralobular pulmonary sequestration with possible congenital pulmonary airway malformation (CPAM), a congenital anomaly known as hybrid lesion.

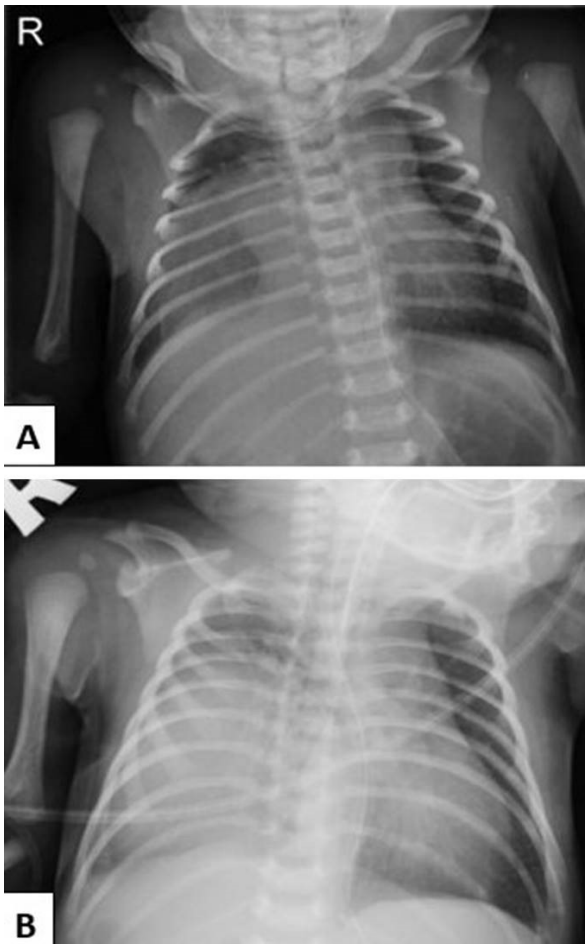

Figure 1: (A) Chest radiograph (PA) acquired at day 1 of life shows opacity at the right mid and lower zones. (B) The subsequent radiograph at day 10 of life shows no significant changes despite completion of antibiotic. (Click on image to enlarge)

The echocardiography showed levocardia, small patent foramen ovale, small inferior vena cava and hypervascularised left hepatic region on subcostal view, suspicious of collaterals from descending aorta. There was also suspicion of partial anomalous pulmonary venous drainage and double aortic arch/aberrant vessel which were unclear from the previous CT. The baby was subsequently referred to our centre for further characterisation of the cardiovascular anomaly, especially the vascular supply and drainage prior to surgery.

Prospective ECG-gated CTA thorax revealed a huge solid mass occupying the entire right hemithorax, sparing its anterior and apical part. The mass extended inferiorly below the hemidiaphragm and insinuated between the liver and right lower posterior ribs. There was an area of non-enhancing hypodensity within it, suggestive of a cystic component. The arterial supply was from the hepatic artery with drainage into the intrahepatic portal vein (Figure 2). No communication existed between the bronchial tree and the mass, suggestive of extralobar sequestration. CPAM could not be excluded. The heart was levocardia with presence of interatrial septal defect (Figure 3). The interventricular septum was intact. There was azygous continuation of the inferior vena cava (image not shown). The aorta and the pulmonary venous return were normal with no evidence of anomalies.

The baby underwent right thoracotomy, right lobectomy and plication of right diaphragm. Intraoperatively, there was abnormal homogeneous fleshy mass of the right lower lobe which adhered firmly to the diaphragm and superior surface of the liver. This mass was separated from the middle lobe

Table I: Differential diagnoses for persistent chest opacity in neonates and the typical radiological findings on chest radiograph and CT scan.

Differential diagnoses	Radiological Findings	
	Chest Radiograph	**CT Scan**
Pulmonary hypoplasia	Soft tissue density mass with ipsilateral mediastinal shift.	Hypoplastic bronchus or pulmonary artery
CPAM (congenital pulmonary airway malformation)	Variable density of the mass depending on the content, with or without air-fluid level. Presence of mediastinal shift.	Mass with cystic component. Presence of pulmonary arterial supply and pulmonary venous drainage.
Pulmonary Sequestration	Soft tissue density mass in the lung base.	Homogeneous soft tissue mass. Presence of systemic arterial supply and abnormal venous drainage.
Bronchogenic Cyst	Water or soft tissue density mass in the mediastinum or lung.	Well-defined cystic lesion. No solid component.
Pleuropulmonary Blastoma	Soft tissue density mass with contralateral mediastinal shift.	Mixed solid cystic mass with contralateral mediastinal shift. Pleural effusion may be present.

with its own covering visceral pleura. Communication with the bronchus was identified but there was no expansion of the lobe observed. The supplying vessels were from the posteromedial part of liver. The right upper and middle lobe showed normal expansion. The liver was eventrated. The intraoperative diagnosis was right extralobar pulmonary sequestration with CPAM. Histopathological examination confirmed CPAM type 2. The baby recovered well post-surgery. To date, the baby is well-thriving post-operative, requiring only 2 admission for acute bronchiolitis. She was given an annual appointment for long-term monitoring.

DISCUSSION

Congenital thoracic anomalies accounts for 2.2 % of livebirth.[1] It can be classified into three categories which are bronchopulmonary, vascular or combined lung and vascular

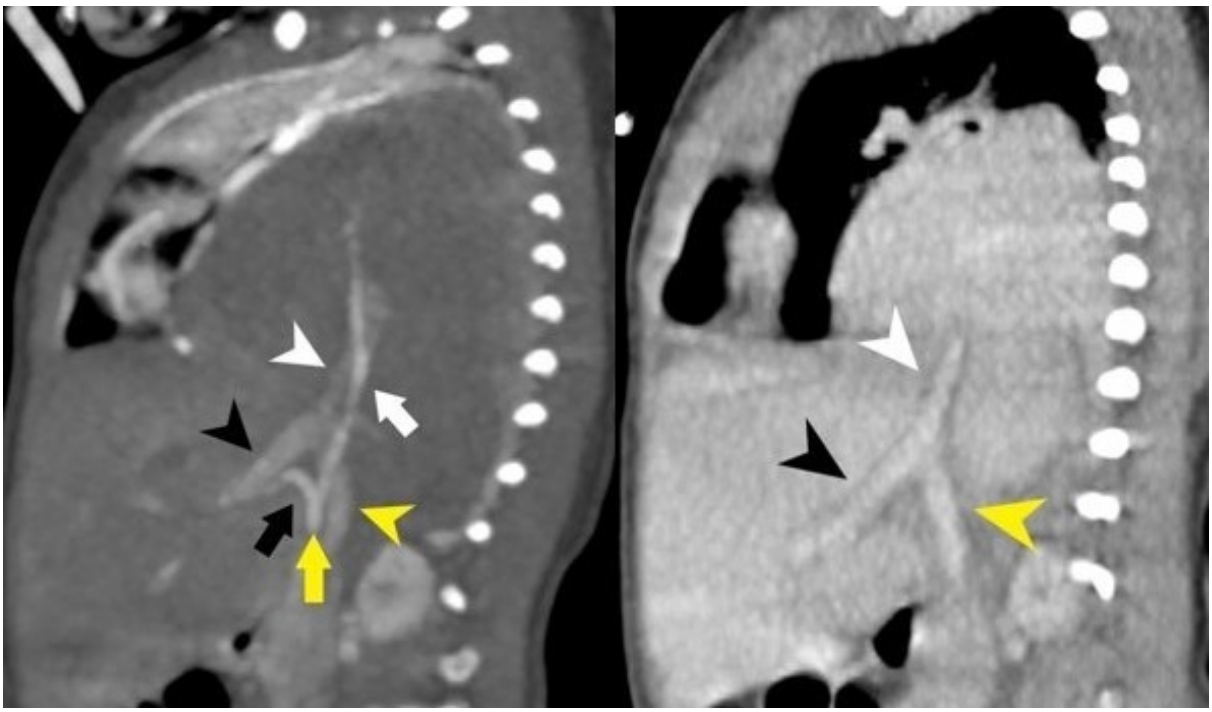

Figure 2: Sagittal reformatted image of CT thorax compare the differentiation of vascular structures in the arterial phase (A) which was performed at our centre and venous phase (B) which was initially performed at primary hospital. The common hepatic artery (yellow arrow) gives rise to branches which supply the liver (black arrow) and the intrathoracic mass (white arrow). The main portal vein (yellow arrowhead) also has branches to the liver (black arrowhead) and to the intrathoracic mass (white arrowhead). (Click on image to enlarge)

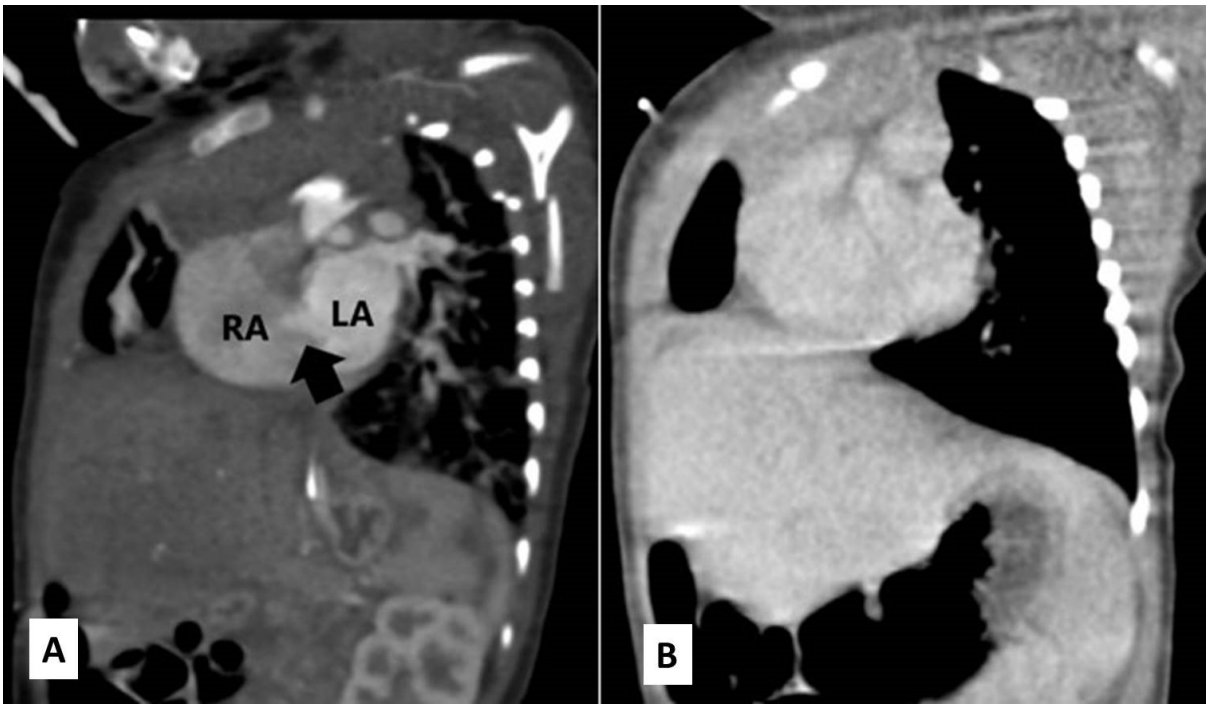

Figure 3: Sagittal reformatted image of CT thorax in arterial (A) and venous phase (B) showing the cardiac anomaly. Note that the interatrial septal defect is appreciated in arterial phase (black arrow) which cannot be appreciated in venous phase. Left atrium (LA) and right atrium (RA) are shown. (Click on image to enlarge)

anomalies (Table II).[2] The most common is congenital pulmonary airway malformations (CPAMs) followed by pulmonary sequestration. These lesions can be diagnosed antenatally by prenatal ultrasonography or MRI.[2]

Table II: Classification of congenital thoracic anomalies and examples.

Classification	Examples
Bronchopulmonary anomalies	• Lung agenesis-hypoplasia complex • Congenital pulmonary airway malformations (CPAMs) • Congenital lobar overinflation (CLO) • Bronchial atresia • Bronchogenic cyst
Vascular anomalies	• Absence of main pulmonary artery • Anomalous origin of the left pulmonary artery or pulmonary sling • Anomalous pulmonary venous drainage • Pulmonary arteriovenous malformations
Combined lung and vascular anomalies	• Scimitar syndrome • Bronchopulmonary sequestration

Antenatally, presence of polyhydramnios or hydrops may have poorer outcome.[6]

CPAMs results from early airway maldevelopment that results from the disorganised proliferation of primary bronchioles.[4,7] It is usually discovered during neonates up to 2 years old; who present with respiratory distress symptoms or recurrent respiratory tract infection. According to Stocker etal., it can be classified into five types (Table III), however, only three types can be differentiated radiologically.[7] These include large cyst CPAM (type I), small cyst CPAM (type II) which are macrocystic, or sol-

Table III: Classification of CPAMs based on Stocker. [4]

Classification	Features
Type 0	Acinar dysgenesis or dysplasia of large airways; incompatible with life
Type 1	Single/multiple large cyst, size >2 cm
Type 2	Multiple smaller cysts <2cm
Type 3	Microcysts <0.5 cm
Type 4	Large cysts of distal acinar (peripheral) origin

id CPAM (type III) which is microcysts.[2] Type II and III CPAMs are associated with other congenital anomalies.[5,6] CPAMs may have abnormal communication with proximal airway, has its own arterial supply and venous drainage from the pulmonary artery and pulmonary vein, respectively. Postnatally, a chest radiograph may show soft tissue opacity or cystic lesion with an air-fluid level which may or may not be associated with the mediastinal shift. It may mimic congenital lobar hyperinflation, loculated pneumothorax, pulmonary sequestration, congenital diaphragmatic hernia or bronchogenic cyst.[4]

Pulmonary sequestration can be classified into intralobar pulmonary sequestration (ILPS) or extralobar pulmonary sequestration (ELPS). Both conditions occurs when a part of the lung does not connect to the tracheobronchial tree and has its own systemic arterial supply, mainly from the thoracic or abdominal aorta.[2] ELPS has its own pleura investment and systemic venous drainage while ILPS shares the pleura investment with normal lung and drains into the pulmonary venous system. EPLS is usually diagnosed neonatally while ILPS is diagnosed later in childhood or adulthood. ELPS is usually associated with other congenital systemic anomalies like pulmonary hypoplasia, cardiac anomalies, congenital diaphragmatic hernia or foregut duplication cyst.[2] Postnatally, a chest radiograph may show soft tissue opacity, usually located at lung bases. It may mimic basal atelectasis or focal eventration of the hemidiaphragm.[4]

In our case, the patient had a hybrid lesion, a combination of both CPAMs and ELPS, which is a rare occurrence. From postnatal radiographs, both CPAMS and ELPS may show similar findings which can only be differentiated by CT, particularly in the hybrid lesion. CTA is better for delineation of anatomical structures, aided by multiplanar reconstruction and volume rendering technique. It is advised to have a scan range from the thoracic inlet to the level of renal artery to ensure adequate coverage.[5] In the paediatric age group, radiation exposure needs to be kept as low as reasonably achievable (ALARA). Therefore, multiphase CT is not necessary since a properly planned CTA is already able to delineate the structures of interest in hybrid lesion.[4]

Ultrasound can be done as the first-line imaging, but it has limited acoustic window as compared with CT.[3] However, the prenatal ultrasound can still serve as a screening tool for prenatal MRI. Since type II CPAMs and ELPS are usually associated with other congenital anomalies, baseline echocardiography should be performed to look for cardiac defects. If the echocardiography findings are equivocal, it is advisable to proceed with a prospective ECG-gated CTA thorax for assessment of lung parenchyma, vascular structure as well as the cardiac anomalies prior to therapeutic decision or surgical intervention. Some centres may perform conventional angiography to identify the arterial supply and venous drainage prior to surgery. The symptomatic patients will need early surgical intervention. However, the management for asymptomatic patients is still controversial.[2,4]

CONCLUSION

Hybrid lesion is a rare congenital thoracic anomaly and understanding of its complex anatomy is important to reach the diagnosis. In an infant with persisting opacity in the hemithorax, CTA is required for accurate anatomical delineation and diagnosis of this condition especially for surgical planning. ECG-gated protocol is to be considered since it can provide differentiation between the vascular structures with less motion artefact. Echocardiography is essential to exclude the associated intracardiac defect.

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CONFLICT OF INTERESTS

I would like to declare that there is no conflict of interest during the completion of this case report.

FINANCIAL SUPPORT

I would like to declare that there is no financial support given in any form.

CONSENT

Verbal informed consent was obtained from the patient's parents for inclusion in this report. Research and ethic committee approval for case report is not a requirement according to the Medical Research and Ethics Committee and Institute for Clinical Research Malaysia.

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