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Inadvertent chest tube insertion in congenital cystic adenomatoid malformation and congenital lobar emphysema-highlighting an important problem

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Abstract

Background: Chest tube insertion in congenital cystic lung lesions is an important problem in children with acute respiratory distress having a cystic lucent lesion on chest radiograph. **Objective:** To evaluate the imaging findings and complications in cases of congenital cystic lung lesions with chest tube insertion and suggest the role of appropriate imaging for management of these patients. **Materials and Methods:** Chest radiographs and CT scans of children with congenital cystic lung lesions who had inadvertent chest tube insertion preoperatively were retrospectively reviewed for imaging appearances and complications. **Results:** Fifteen patients comprising 10 cases of congenital cystic adenomatoid malformation (CCAM) and 5 cases of congenital lobar emphysema (CLE) were included. Majority of the cases were infants. CCAM was misdiagnosed as complicated pneumatocele ($n = 5$) and pneumothorax ($n = 5$), while CLE was misdiagnosed as tension pneumothorax ($n = 5$) on the chest radiograph findings. Final diagnosis was made on CT and operative findings with histopathology. Complications noted were pneumothorax, hydropneumothorax, and infection in cases of CCAM, and change in imaging appearance and pneumothorax in cases of CLE. **Conclusion:** Chest tube insertion in congenital cystic lesions increases the rate of associated complications. Chest CT has a definite role in early diagnosis and deciding appropriate management in these cases.

Key words: Chest tube insertion; cases of congenital cystic adenomatoid malformation; congenital lobar emphysema; cystic lucent lesion; pneumatocele; pneumothorax

Introduction

Cystic lucent lung lesion on chest radiograph is a common finding in children presenting with acute respiratory distress. The prominent differentials for cystic lucent chest lesion on chest radiograph in infants and young children (under 3 years of age) with respiratory distress are post-infective pneumatocele, pneumothorax, congenital diaphragmatic hernia (CDH), congenital cystic adenomatoid malformation (CCAM), congenital lobar emphysema (CLE), pulmonary sequestration, bronchogenic cyst, and bullous lung disease.[1-3] Congenital cystic lung lesions are relatively rare in incidence and have a varied clinical presentation.[4] Majority of the patients usually present in infancy or within the first 3 years of life. On chest radiograph, these patients usually present as cystic lucent lung lesions with or without mass effect. In developing countries, the incidence of chest

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infection and infection-related complications (pneumothorax and pneumatoceles) is high, and they are considered to be the more common causes of lucent cystic lesions on chest radiograph in patients with respiratory distress.[5] Chest tube insertion into congenital cystic lung lesions, mistaking them for an infection-related complication, is a recurring clinical problem which has been reported in the literature as isolated case reports/series and is associated with increased rate of complications in the patient [Table 1].[6-12] The major errors in diagnosis leading to chest tube insertion are due to the inability to differentiate CCAM from tension pneumothorax/pneumatocele and CLE from a tension pneumothorax. The purpose of the study was to evaluate the imaging findings and complications associated with chest tube insertion in congenital cystic lung lesions and review the role of imaging for appropriate management of these patients. In addition, we aimed to highlight this important recurring problem.

Materials and Methods

We reviewed the case records of children with congenital cystic lung malformations who were operated at our institution from 1998 to 2011. Patients with congenital cystic lung malformation confirmed on imaging (chest radiograph and CT), surgery, and histopathology, and who had an inadvertent chest tube insertion preoperatively were included in the study. We collected information related to clinical presentation, imaging findings on chest radiograph and CT, and operative and histopathology findings. An investigator studied the imaging findings on plain chest radiograph (PA or AP) before and after tube insertion. CT scan was performed with a 40-slice multi-detector computerized tomography (MDCT) scanner (Philips Brilliance) using a routine chest CT protocol for children, with 80-kVp tube voltage and 80-mA tube current. Intravenous iodinated contrast medium (low osmolar, non-ionic, 300 mg/ml iodine content) was used routinely at a dose of 2 ml/kg body weight, administered by hand injection. The mean CT dose index volume in our study was found to be 3.0 ± 0.8 mGy.

The imaging findings at CT were correlated with chest radiograph and surgical findings. We also studied the CT images for changes in imaging appearance and complications due to chest tube insertion.

Results

Out of 54 children operated for congenital cystic lung lesion, 15 children (27%) with an inadvertent chest tube insertion preoperatively based on plain radiograph findings were identified. The children were categorized into two groups based on the final diagnosis as CCAM and CLE.

Cases diagnosed as CCAM

There were 10 cases (66%) of CCAM in the series [Table 2]. Respiratory distress was present in all 10 patients at presentation, fever in 3 patients, cough in 3 patients, and cyanosis in 1 patient. A prenatal ultrasound (US) study had not been performed in any of the patients in the study. Chest radiograph findings revealed cystic hyperlucent lung lesion in all cases. Multiseptated cystic hyperlucent lesion was seen in nine cases, while one case showed a single cystic lucency with no septations. The cyst size was uniformly the same in two cases, while in seven cases there were variable-sized cystic lucencies with a single large dominant cystic lucency. In seven cases, the cystic hyperlucent lesion was seen localized to a lung segment, while in three cases it was seen to involve the whole lung field. Mass effect

Table 1: Chest tube insertion in CCAM and CLE: Review of literature

Author (ref.)	Final diagnosis	No of cases	Diagnosis mistaken as	Age of patient	Site of lesion
Nandi (6)	CCAM	1	Pneumothorax	1 month	-
	CLE	1	Pneumothorax	8 months	LUL
Powers (7)	CLE	1	Tension pneumothorax	23 months	RUL
Kumar (8)	CLE	2	Pneumothorax	-	-
	CCAM	1	Pneumothorax	-	-
Tempe (9)	CLE	3	Tension pneumothorax	1 month	LUL
				2 months	LUL
				1 month	RUL
Ulku (10)	CLE	3	Tension pneumothorax	12 months	RUL
				2 months	RLL
				NB	RUL
Sittig (11)	CCAM	1	Pneumothorax	NB	Left lung

CLE: Congenital lobar emphysema, CCAM: Congenital cystic adenomatoid malformation

Table 2: Summary of cases with complications

Final diagnosis	Age	Sex	Site of lesion	Diagnosis mistaken as	Complications due to tube insertion
CCAM	18 days	F	RLL	Pneumothorax	-
CCAM	3 months	M	LLL	Pneumatocele	Hydropneumothorax, subcutaneous emphysema
CCAM	8 months	M	RUL	Pneumatocele	Pneumothorax, subcutaneous emphysema
CCAM	2 months	M	RUL	Pneumothorax	Infection, pneumothorax
CLE	8 months	M	RUL	Pneumothorax	Pneumothorax, subcutaneous emphysema
CLE	1.5months	M	LUL	Pneumothorax	-
CLE	1 month	M	LUL	Pneumothorax	Pneumothorax, subcutaneous emphysema
CLE	9 months	M	LUL	Pneumothorax	-
CCAM	14 months	M	RLL	Pneumatocele	Infection
CCAM	3 months	M	LLL	Pneumatocele	
CLE	7 months	M	LUL	Pneumothorax	Pneumothorax, subcutaneous emphysema
CCAM	3 years	M	RUL	Pneumatocele	Infection
CCAM	3.5 months	M	RLL	Pneumothorax	-
CCAM	5 months	F	RUL	Pneumothorax	Pleural effusion, infection
CCAM	4 months	M	LUL	Pneumothorax	Hydropneumothorax, infection

CLE: Congenital lobar emphysema, CCAM: Congenital cystic adenomatoid malformation

with resultant mediastinal shift was seen in seven cases, while three cases showed cystic lucent lesion with no mass effect. Following chest tube insertion, respiratory distress increased in all 10 patients and a CT scan was performed to rule out complications. Worsening of respiratory distress was determined based on clinical notes; in some cases, objective measures (increased oxygen requirements) were available.

On CT, multiple thin-walled cystic lesions involving an entire lobe of lung were seen. Multiseptated cystic lesions with single dominant cyst were seen in nine cases, while single air-filled cystic lesion was seen in one case. The diagnosis was type 1 CCAM in nine cases and type 2 CCAM in one case. The affected lung segment showed no normal intervening lung parenchyma between the cysts in any of the cases. Mass effect and mediastinal shift were seen in seven cases. Of the three cases in which the cystic lucency was seen involving the whole lung field on plain radiograph, CT showed the lesion to lie in the left lower lobe in two cases and in the right lower lobe in one case**.** One of the cases who did not have mass effect on initial chest radiograph showed mediastinal shift on CT following chest tube insertion. Due to chest tube insertion, moderate hydropneumothorax and pneumothorax were seen in two cases each, resulting in some volume loss of the surrounding lung. Pleural effusion was noted in one case and subcutaneous emphysema in two cases. In four cases, air fluid levels and consolidation developed following chest tube insertion, which was not present on the initial radiograph, which suggested the development of secondary infection. This was correlated with clinical findings and blood counts. All 10 patients underwent lobectomy and diagnosis was confirmed at histopathology. As a result of the complications due to chest tube insertion, adhesions were noted around the cyst that caused difficulty during surgical excision of the lesions. No mortality was noted as a result of chest tube insertion.

Cases diagnosed as CLE

There were five cases of CLE misdiagnosed as pneumothorax on initial plain radiograph with resultant chest tube insertion in the study. A prenatal US study had not been performed in any of the patients in the study. All patients had dyspnea at presentation, but only three patients were in severe respiratory distress. Chest radiograph was diagnostic in four cases, showing hyperinflated lung field herniating across the midline with associated mediastinal shift. Faint vessel markings were seen in all cases within the hyperlucent lung field. In one case, however, the hyperinflated lung field was not obvious on the initial radiograph because of prior chest tube insertion. Due to non-improvement of patient's condition, the chest tube was repositioned, but the patient's respiratory distress worsened. In all five cases, the diagnosis was mistaken as tension pneumothorax on initial chest radiograph with resultant chest tube insertion. Due to worsening of the patient's dyspnea following tube insertion, CT scan was performed. CT was diagnostic in four cases of CLE, showing hyperinflated affected lobe with attenuated vascular markings and herniation of lung across the midline with mediastinal shift. In one case of CLE, multiple chest tube insertions resulted in consolidation, pulmonary hemorrhage, pneumothorax, and led to loss of typical diagnostic hyperinflated lung segment on CT. This made it difficult to make a definite diagnosis of CLE on the initial CT. However, despite the chest tube insertion, the affected lung segment looked mildly hyperlucent compared to the surrounding lung and a possibility of CLE was suggested. The patient's chest tube was removed, which improved the respiratory distress. A repeat CT of the patient 3 months later showed the classical hyperinflated

lobe suggestive of CLE. Due to chest tube insertion, subcutaneous emphysema was seen in three patients and moderate pneumothorax in three patients resulting in minimal volume loss of the adjacent lung. All five patients were operated and diagnosis confirmed on histopathology. No mortality was noted as a result of chest tube insertion.

Discussion

CCAM is a developmental abnormality of lung in which there is abnormal adenomatoid proliferation of bronchial structures as cysts rather than normal alveoli and is limited to a single lobe in the majority of cases. These abnormalities usually present in neonates and infants, but are also known to occur in adults. The majority of patients with CCAM in our series were infants. Stocker *et al*. classified CCAM into three types depending upon the size of cysts contained in the lesion.[13] The appearance of the lesion on chest radiograph and CT depends upon the number, size, and amount of fluid within the lesion. On plain radiograph, CCAM can present as a cystic lucent lung lesion with or without mass effect. There are multiple isolated reports of chest tube insertion in CCAM, after mistaking it for complicated pneumatocele and tension pneumothorax.[6,8,11] In our study, CCAM was mistaken as pneumatocele in five cases and pneumothorax in five cases on initial chest radiograph, resulting in chest tube insertion. In seven cases, cystic lucency was localized to a part of the lung field either in the upper or lower lobe, while in three cases of CCAM involving the lower lobes, it was seen to extend over the entire lung field [Figure 1]. Thus, CCAM involving the lower lobes might resemble a pneumothorax by showing lucency across the entire lung field; however, the presence of septations within the cystic lucent lesion, presence of vascular markings, and relative lack of lucency in the uninvolved apical region point toward a diagnosis of CCAM. A multiseptated cystic lucent lesion was seen in nine cases, while single cystic lucency was noted in only one case. Thus, the presence of septations within a cystic lucent lesion should suggest a possibility of CCAM. Mass effect and the resultant mediastinal shift were seen in the majority of our cases.

As noted in our study, CT was diagnostic in all cases, and helped in correctly localizing the lesion, detecting the position of chest tube, and demonstration of complications due to chest tube insertion. Thus, it had an additive role over chest radiograph. The complications noted in our series were pneumothorax, hydropneumothorax, pleural effusion, infection, and subcutaneous emphysema [Figures 2-4] [Table 2]. In addition, tube insertion resulted in the formation of adhesions around the cyst due to infection which caused difficulty during surgical excision.

In our series, 50% cases of CCAM were misdiagnosed for complicated pneumatoceles. Pneumatoceles are post-infectious thin-walled collections of air within the lung parenchyma, commonly seen in infants and children up to 3 years of age, and are generally known to appear during the resolving stages of staphylococcal and streptococcal pneumonitis. However, Kunyoshi *et al.* noted that pneumatoceles may be seen on the first chest radiograph in 80% of the cases and that up to 90% of the pneumatoceles show spontaneous resolution by 3 months.[5] The imaging findings of CCAM may be indistinguishable from those of pneumatoceles in some cases on chest radiograph and CT.[14] The presence of normal lung parenchyma between the cysts may be an important CT criterion to differentiate a pneumatocele from a CCAM in which the entire affected part of lung parenchyma is abnormal. Although in developing countries pneumothorax/pneumatocele may be more common causes of cystic chest lucency, a possibility of a congenital cystic malformation must be always be kept in mind, especially in children less than 3 years of age, when multiple cystic lucencies are seen in a localized area with no surrounding consolidation. There is paucity of literature regarding the ability of plain radiograph and CT to correctly differentiate CCAM from pneumatoceles, and further studies may be required. Chest tube insertion should be avoided in congenital cystic lung lesions, and a prior CT is advocated if atypical findings are found on the chest radiograph. Direct chest tube insertion is recommended only in extreme cases with severe respiratory distress and

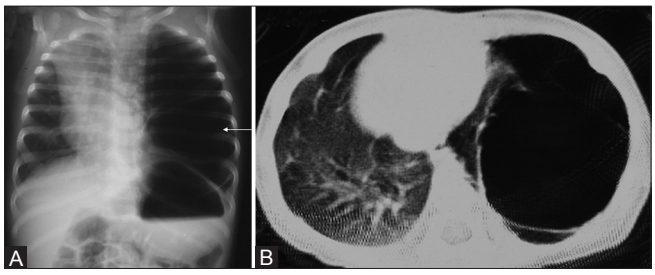

Figure 1 (A, B): A 3-month-old infant with respiratory distress. (A) Chest radiograph shows cystic hyperlucent left lung field with septations (arrow) and mass effect. This was mistaken as pneumatocele with resultant chest tube insertion. (B) CT chest axial section with lung window shows a thin-walled cyst in left lower lobe with mass effect. Diagnosis was confirmed after surgery as type 1 CCAM

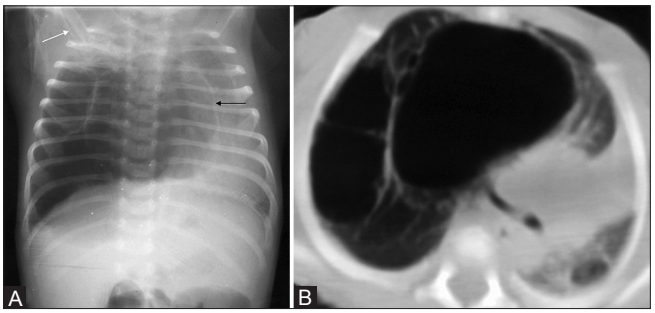

Figure 2 (A, B): Infant with respiratory distress. (A) Chest radiograph shows a multiseptated cystic lucency (black arrow) in the right lung field with mediastinal shift. A chest tube (white arrow) was inserted after mistaking it for a pneumothorax. (B) CT chest axial section with lung window shows multiple thin-walled cysts in the right upper lobe. Diagnosis was confirmed after surgery as type 1 CCAM

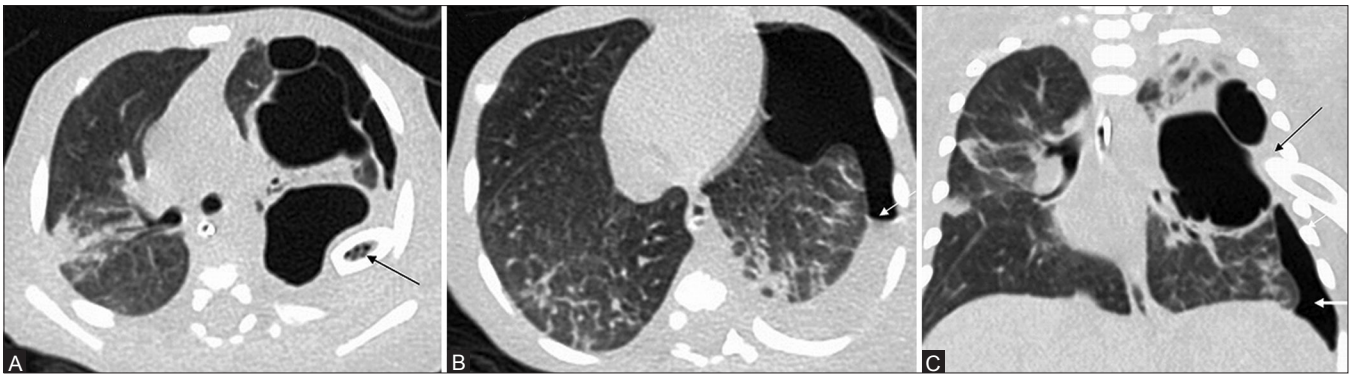

Figure 3 (A-C): CT chest lung window axial sections show (A) chest tube insertion (black arrow) in a multiseptated cystic lesion in the right upper lobe, (B) resulting in the formation of hydropneumothorax (white arrow). (C) Coronal image shows the chest tube (black arrow) in the multicystic lesion and the resultant pneumothorax (white arrow). Diagnosis was confirmed after surgery as type 1 CCAM

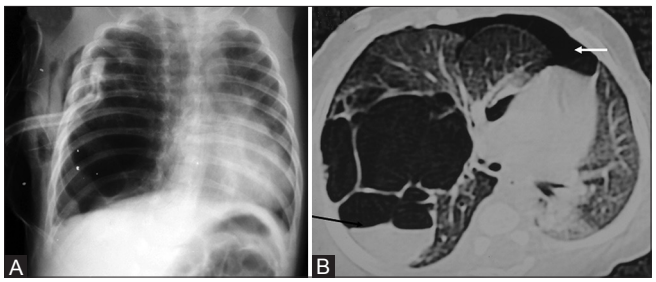

Figure 4 (A, B): (A) Chest radiograph shows cystic lucency involving the whole of the right lung field with chest tube insertion and subcutaneous emphysema. (B) Chest CT axial section with lung window shows multiple thin-walled cystic lesions showing air fluid level (black arrow) and pneumothorax (white arrow). Diagnosis was confirmed after surgery as type 1 CCAM

significant mediastinal shift which may decompress a potential pneumothorax, tension pneumatocele, or a large CCAM. This also forms the basis of fetal thoracoamniotic shunting for decompression of large CCAM.[15,16] Also, as noted in our study, no increased mortality was seen due to chest tube insertion.

CLE is a rare congenital lung anomaly characterized by postnatal overexpansion of one or more lobes of a histologically normal lung. The most common lobes affected are left upper lobe followed by right middle and right upper lobes.[2] The left upper lobe (four cases) was the most commonly affected lobe in our study. Majority of the patients present in the immediate neonatal period; however, presentation in late infancy and early childhood is well known.[2] On chest radiograph, a hyperlucent, hyperexpanded affected lung field is seen that shows herniation across the midline and mediastinal shift.[3] This radiographic appearance can be mistaken for a pneumothorax/tension pneumothorax with resultant chest tube insertion, and this problem has been widely reported in the literature.[6,7,9,10] Chest tube insertion in CLE worsens the patient's condition by producing pneumothorax, subcutaneous emphysema, and increase in mediastinal shift, and may result in delay in diagnosis as was noted in our study. This is, however, clearly avoidable by careful clinical and radiological correlation. Careful observation of the chest radiographs in CLE will demonstrate scant lung markings within the hyperlucent area, which will not be seen in case of a pneumothorax [Figure 5]. All the cases in our study showed faint vascular markings within the hyperlucent area. This finding is, however, subject to interpretive error as evidenced by multiple case reports reporting this problem. In addition, in CLE compression of the adjacent lobes pushes them cephalad or caudal toward the diaphragm, while in cases of pneumothorax, the lung collapses toward the hilum. Similar observations have been noted in previous reports.[10] CT is diagnostic in CLE, and is recommended for confirmation for diagnosis and for delineation of the affected lobe preoperatively. On CT, hyperinflated lobe with attenuated and displaced pulmonary vessels is a typical appearance of CLE.[2] However, as seen in one of our cases, with multiple chest tube insertions this typical imaging appearance may be lost making a definitive diagnosis difficult even at CT [Figure 6].

Chest radiograph is an important initial investigation in the evaluation of cystic lucent lung lesions. Appropriate differentials may be considered by noting the size, position, location, number, and evolution of cysts, the age of patient, and by comparison with previous chest radiographs.[1] Detailed chest radiograph evaluation of the lung borders, bronchovascular markings, mediastinal shift, septations within cysts, consolidation surrounding the cystic lucency, position of collapsed lobes, diaphragmatic margins, and review of previous X-rays in an emergency setting is vital for correct diagnosis. However, the imaging findings of CCAM may be indistinguishable from those of pneumatoceles in some cases on chest radiograph. US is a sensitive modality for the detection of pneumothorax.[17,18] The absence of comet tail artifacts and sliding lung sign is indicative of air in the pleural cavity, and US may be used as an initial screening modality in differentiating a CLE from a tension pneumothorax. CCAM may be demonstrated on US as

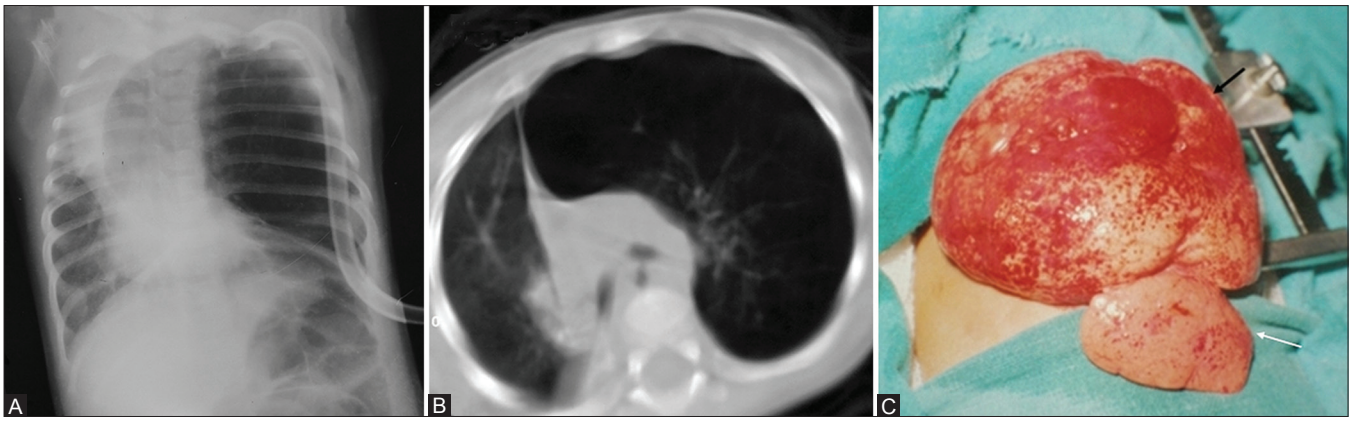

Figure 5 (A-C): (A) Chest radiograph showing hyperlucent left lung field herniating across the midline with preserved vascular markings. This was mistaken as pneumothorax with resultant chest tube insertion. (B) CT axial section lung window shows a hyperinflated left lung segment with attenuated lung markings. (C) Peroperative image shows the hyperinflated left upper lobe (black arrow) in comparison to the normally aerated left lower lobe (white arrow). Diagnosis after surgery confirmed this as a left upper lobe CLE

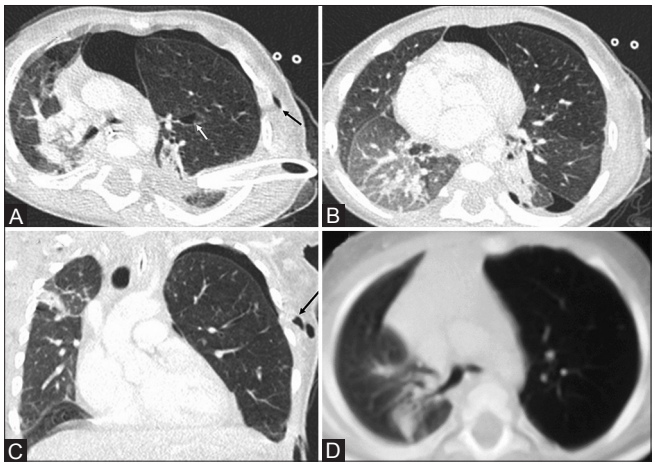

Figure 6 (A-D): A 7-month-old child with multiple chest tube insertions due to clinical suspicion of pneumothorax. Chest CT images lung window axial and coronal views (A-C) show a slightly hyperinflated left upper lobe with a pneumothorax due to multiple chest tube insertions (black and white arrows). However, on comparison with adjacent lung segments, the hyperinflation is not obvious for definitive diagnosis of CLE. (D) Follow-up CT 3 months later shows hyperinflated left upper lobe with attenuated vascular markings, diagnostic for CLE. Diagnosis was confirmed after surgery

multiple cystic anechoic to homogenously solid hyperechoic lesions within the lung parenchyma.[19,20] However, differentiation of large cystic CCAM from pneumatocele and pneumothorax may be difficult on US and further studies are required.[18] Chest CT has a definite role in the management of children with respiratory distress and cystic lucent lung lesion on chest radiograph, as was noted in our study. CT will help in confirming the diagnosis and provides vital information for operative and image-guided intervention.

Chest CT is not recommended in all cases of suspected pneumothorax. It is only recommended when the imaging findings are atypical and respiratory distress does not improve or worsens following chest tube insertion, which would not be the case in a simple pneumothorax. Role of CT in such cases would be to diagnose alternate conditions such as large CCAM/CLE, which may mimic pneumothorax/pneumatocele on chest radiograph. In addition, it may help in detecting complications associated with chest tube insertion, which can help explain patient's symptoms. Due to the retrospective nature and criteria for inclusion for the study, we could not opine in how many cases chest CT would have prevented a chest tube insertion. However, in all cases CT provided a diagnosis of congenital lung abnormality and this resulted in removal of the chest tube. Thus, this made us conclude that a prior CT would have prevented a chest tube insertion.

A potential limitation of our study was that we did not include cases of complicated pneumatocele for imaging comparison. Also, we did not have CT scans of patients before chest tube insertion and it was assumed that the complications were a result of the chest tube insertion based on clinical and imaging findings on initial chest radiograph. Due to the retrospective nature of our study over a long duration, the exact details of the training level of the primary reader of the chest radiograph were not available. In addition, the criteria for inclusion in our study (chest tube insertion in patients with congenital lung lesions) probably resulted in inclusion of cases with relatively atypical findings on chest radiograph.

Conclusion

Chest tube insertion in congenital cystic lung lesions worsens the respiratory distress and increases the rate of complications like pneumothorax, hydropneumothorax, infection, and subcutaneous emphysema. It may also result in change in imaging findings, resulting in delay in diagnosis. Chest CT has an important role in confirming the diagnosis, detecting the complications, and deciding appropriate therapy.

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