Congenital Pulmonary Adenomatoid Malformation in three Infants with Diagnostic and Management Challenges in a Low and Middle-Income Country: A Case Series

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ABSTRACT

Congenital Pulmonary Adenomatoid Malformation (CPAM)is a rare disorder consisting of harmatomatous and dysplastic lung tissues. Few cases have been reported in Nigeria.We report CPAM in three infants, to highlight the occurrence of this rare condition in our locality, raise awareness for high index of suspicion, and explore regional management challenges. The first patient, a 6-month old female, presented in March 2016, with complaints of recurrent cough, fast breathing and weight loss of 3 months duration. She was initially diagnosed with lobar pneumonia. Computerized tomographic (CT) scan of the chest showed features in keeping with CPAM (type 1). In April 2019 a 12 day old male presented with high grade fever, cough and fast breathing. Chest radiograph revealed pneumonic changes and macrocystic lesions in the right mid and lower lung zones. Chest CT showed thick and thin-walled, rounded, cystic areas of varying sizes within the right hemithorax, suggestive of CPAM. In June 2019, a 3 month old female presented with a five day history of fever and cough and a 4 day history of fast breathing. An initial diagnosis of pneumonia with sepsis was made and the patient was commenced on parenteral antibiotics. Chest CT revealed features highly suggestive of CPAM. Patients' acute symptoms resolved following medical management. All the parents declined surgery.The possibility of an underlying congenital lung anomaly though considered rare should be explored when children present with recurrent respiratory tract infections or show poor response to treatment for respiratory tract infections.

Keywords: CPAM, Ignorance, Poverty, Cystic, Recurrent, Pneumonia

BACKGROUND

Congenital lung anomalies (CLA) are a heterogenous group of developmental disorders of the lungs and exist in various forms.[1,2] These include pulmonary vascular anomalies like pulmonary agenesis-hypoplasia complex and parenchymal anomalies like congenital large hyperlucent lobe and CPAM.[1,2] Over the years, theories on causation have ranged from defects in development at the pseudoglandular phase of lung development, to genetic signal

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Specialty Section:
This article was submitted to Paediatrics, a section of TJMR.

***Received:** 27 February 2022*
***Accepted:** 21 April 2022*
***Published:** 23 July 2022*

Citation:
Ndukwu C, Elo-Ilo JC, Ugwu NO, Ofiaeli OC, Umeh EO, Mbachu CNP, et al., Congenital Pulmonary Adenomatoid Malformation in three Infants with Diagnostic and Management Challenges in a Low and Middle-Income Country: A Case Series. Trop J Med Res. 2022:21(1);235-245. DOI: 10.5281/zenodo.6890674

Access Code

http://tjmr.org.ng

pathway aberrations. [2,3,4] The commonest type reported has been the congenital pulmonary airway malformation (CPAM) in 30 to 40% of cases of CLA.[2]

CPAM is a rare disorder consisting of harmatomatous and dysplastic lung tissues which is usually confined to one lobe of the lungs.[5-7] It develops during the branching and proliferation of the bronchial structures.[6] There are different forms with the most lethal developing from the tracheobronchial tree. [6] The disorder has an estimated incidence of approximately 14 per 100,000 live births, or 1:11,000 to 1:35,000 pregnancies using prenatal ultrasounds.[2,5-8] The first reports of Congenital Adenomatoid Malformations of the lung appeared in literature over 70 years ago.[9] It was initially classified into 3 types by Stocker et al; Type 1- macrocystic and consists of single or several large cysts (>2 cm in diameter), this makes up 50% of the cases, type 2 (40%) is microcystic consisting of multiple small cysts and it is usually associated with other congenital anomalies with poorer prognosis.[3] Type 3 (< 10 %) are usually solid with bronchiole like structures. This carries the poorest prognosis. This classification was subsequently revised to 5 subtypes based on the tracheobronchial or acinar structures involved .[10,11]

Aside from prenatal diagnosis using ultrasound which is the mainstay for diagnosis of CPAM in the developed world, some cases of CPAM are diagnosed in neonates, infants and young children and even adults.[11,12] Postnatal diagnosis is frequently confirmed using Computerized Tomographic (CT) scan. [6,7] Histology of tissue biopsies further consolidates and reaffirms the pathologic diagnosis.[6,7]

There are very few reports of this anomaly in Nigeria with none in the South Eastern region. The two reported in South West Nigeria were initially misdiagnosed with the right diagnosis made following imaging studies.[13,14] We therefore report the cases of three infants consecutively managed over a period spanning 4 years, with clinical and radiologic features consistent with CPAM to highlight the occurrence of this rare condition in our region and explore diagnostic and regional treatment challenges.

CASE PRESENTATION

CASE 1: A 6-month old female, only child of parents, presented to the Children Emergency Room of the Hospital, in March 2016, with complaints of recurrent fast breathing noticed about a week after birth, recurrent cough, and weight loss of 3 months duration. Pregnancy and birth history were normal. Her feeding was adversely compromised during periods of fast breathing. Over the course of the illness, she received treatment from primary health facilities for recurrent respiratory tract infection (RTI).

She was febrile, tachypnoeic, with a weight of 4.6kg which was 61% of expected. Chest percussion was dull and breath sounds were markedly reduced on the left hemithorax. Abdominal and genitourinary exams were normal. The clinical diagnosis was left sided lobar pneumonia with Protein Energy Malnutrition (PEM). She had a white blood cell count of $13.6x10^6$/L (Neutrophils 61%, lymphocytes 39 %) and the blood film showed toxic granulations. Mantoux skin test and retroviral screening were negative. Chest X-ray showed multiple thick walled cystic lesions within the left hemithorax. Computerized Tomographic scan of the chest showed multiple, well defined, rounded air dense lesions of varying sizes measuring 2-21mm seen within the left hemithorax with associated ring enhancing, likely fluid-filled areas of intermediate density (HU 14), findings consistent with CPAM (possibly type 1). See Figure 1. She was commenced on antibiotics and nutritional rehabilitation. Following cardiothoracic surgery review, she was booked for lobectomy, but parents declined consent on the grounds of financial

constraints and fear of a possible negative outcome. Patient's condition remained relatively stable with decreasing recurrence of respiratory symptoms. Her weight was optimal at last follow up, being 12kg at 2 years.

CASE 2: A 12 day old male presented at the Children Emergency room in April 2019, with a 10 day history of high grade fever, a week history of cough associated with post tussive vomiting and 3 days history of fast breathing which was so severe that the child could not suckle at the breasts. Fast breathing was initially noticed a few hours after birth but resolved shortly after he was given an unknown intramuscular injection. Occasional bluish discoloration of the lips and extremities was observed before presentation. His only elder sibling had died at 2 weeks of age from a febrile illness associated with fast breathing and convulsion.

At presentation, patient was lethargic, in severe respiratory distress, tachypnoeic, grunting with an oxygen saturation of 66% with room air. He had tachycardia. The right hemithorax was dull to percussion, with markedly reduced breath sounds. He had bibasal crepitations. An initial diagnosis of neonatal sepsis with pneumonia was made. The child received intravenous antibiotics and intranasal oxygen for 60 hours.

Full blood count showed a relative neutrophilia and he had an ESR of 16mm in the 1st hour. Chest radiograph showed a homogenous opacity within the right lung apex and inhomogenous opacities within the peripheral aspect of the ipsilateral mid zone suggestive of pneumonic changes. Also, large rounded air-lucent areas were appreciated within the right middle and lower zones devoid of lung markings, likely cystic and suggestive of a congenital pulmonary anomaly. Chest ultrasound showed air induced areas of acoustic shadowing over both lungs degrading image sensitivity and was inconclusive. Chest CT done confirmed presence of multiple, thick and thin-walled, rounded, cystic areas of varying sizes within the right hemithorax predominantly involving the middle and lower lobes, with appearances suggestive of CPAM. (Figure 2) The patient recovered from the respiratory tract infection but remained for about a month in the hospital because the parents were unable to pay their bills at the time of discharge. The need for subsequent surgery was communicated to parents who declined, citing financial difficulties as their major constraint. They have since not returned.

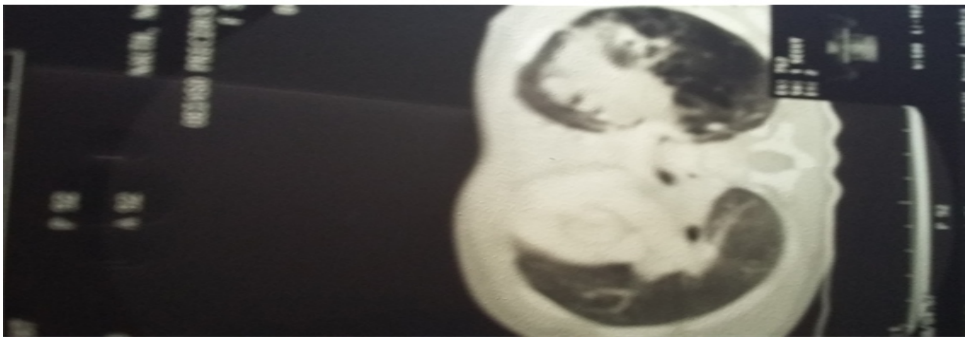
CASE 3: A 3 month old female presented to the children emergency room of Nnamdi Azikiwe University Teaching Hospital (NAUTH), Nnewi, Nigeria in June 2019, with a 5-day history of fever and cough and a 4 day history of fast breathing. She was completely vaccinated within the recommendation for her age group. Pregnancy and birth history were normal and she was the only child of the parents at the time.

On examination, patient was noted to be in marked respiratory distress, gasping for air with an oxygen saturation of 81%. Patient was mildly febrile with a temperature of 37.7^0 C. Clinical findings on the chest included dull percussion notes with decreased breath sounds, restricted to the right hemi-thorax. Her liver was enlarged 6cm below the right costal margin. The digestive and genitourinary system examinationwas essentially normal.

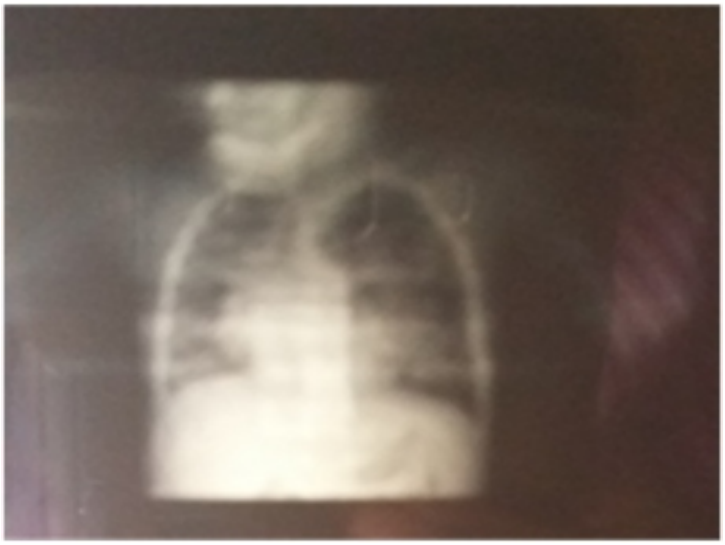
Investigations conducted at the time of presentation included a full blood count which showed absolute leucocytosis and thrombocytosis. Retroviral screening was negative and Serum electrolytes, Urea and creatinine were normal. An initial diagnosis of pneumonia with sepsis was made and the patient was commenced on parenteral antibiotics. Other laboratory and imaging tests were delayed because of financial constraints.

The chest radiograph retrieved a week after admission, revealed a fairly homogenous opacity

(1a)

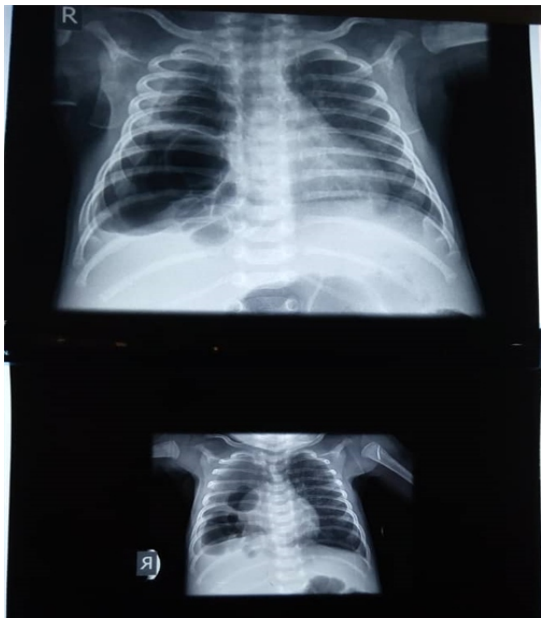


(1b)

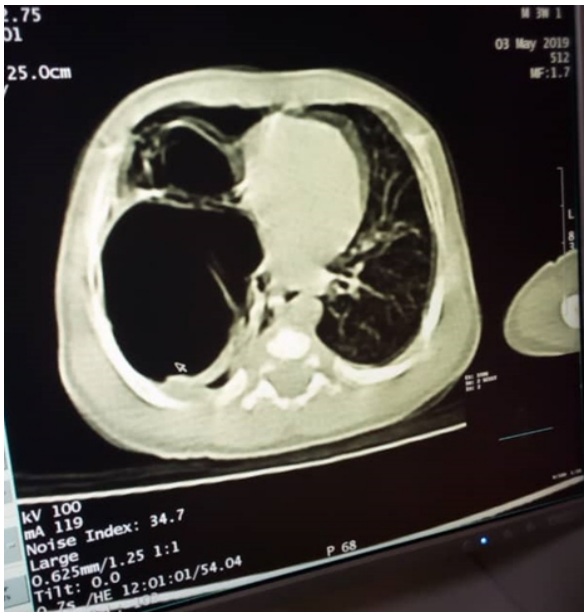


(1c)

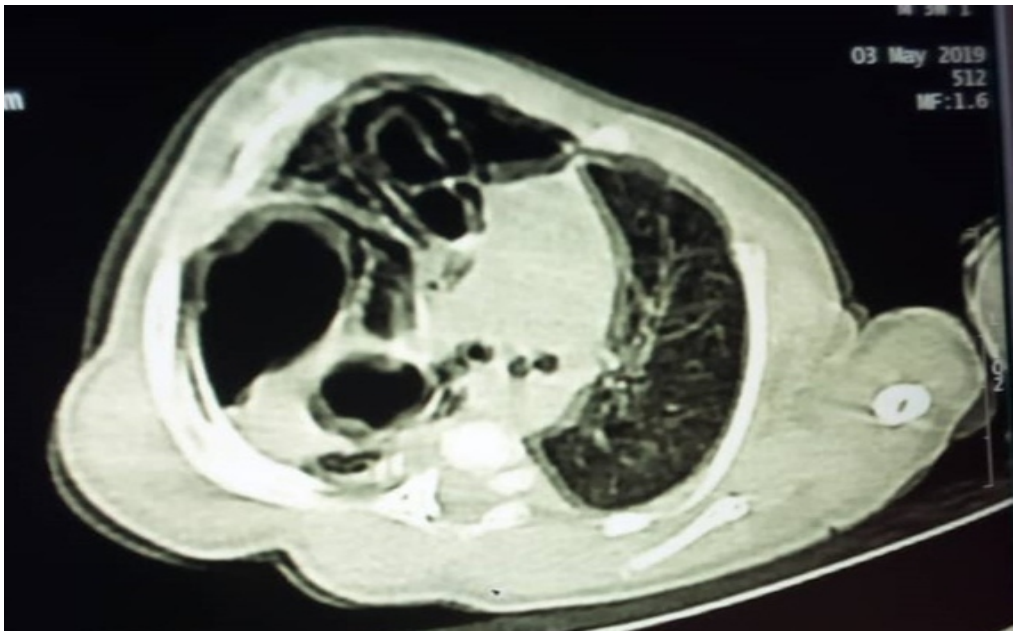
Figure 1: (a, c) Computed tomographic scan of the chest showing multiple, well defined, rounded air dense lesions of varying sizes within the left hemithorax with associated ring enhancing. (b) Chest radiograph revealing multiple thick walled cystic lesions within the left hemithorax.



(2a)

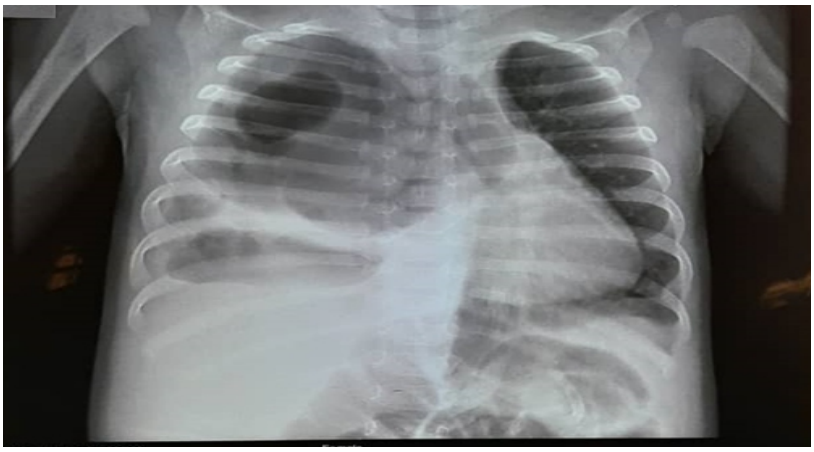


(2b)

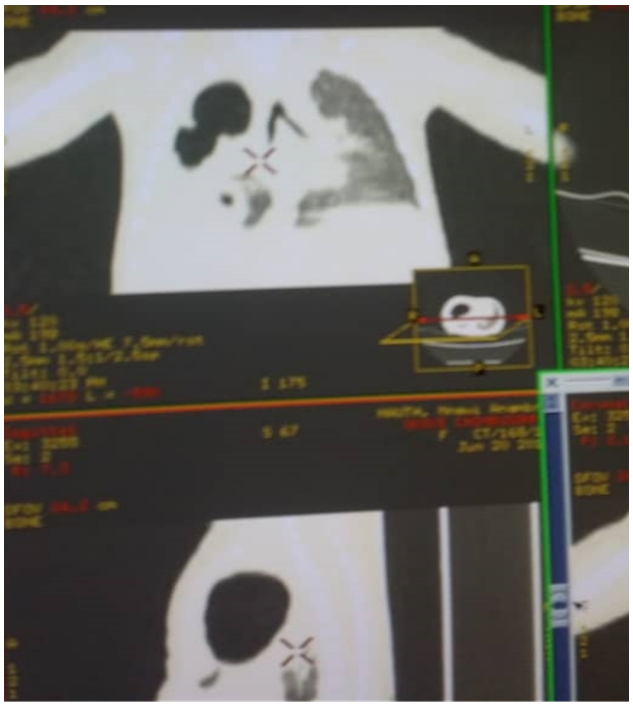


(2c)

Figure 2: (a) Chest radiograph revealing homogenous opacity within the right upper lobe and scattered inhomogenous opacities within the middle lobes with associated macrocystic areas.(b,c) Computed tomographic scan of the chest showing irregular thick walled, air filled cystic lesions



(3a)



(3b)

Figure 3: (a) Chest radiograph revealing homogenous opacity within the right lower lobe obliterating the ipsilateral costophrenic sulcus and a curvilinear density traversing the right mid zone with an air lucency at the right upper zone devoid of lung markings with an increase in right lung volume with some extension across the midline to the contralateral side. (b) Coronal computed tomographic scan of the chest showing an irregular air filled macrocystic lesion.

within the right lower and mid zones, extending to the right lung apex and also obliterating the ipsilateral costophrenic sulcus giving the so-called 'ground glass' features. A curvilinear density was noted traversing the right mid zone likely due to thickened horizontal fissure with pleural fluid or fibrous changes. Overlapping, rounded air lucencies are seen projected over the right upper zone and devoid of lung markings. An increase in right lung volume was noted, with some extension of the medial border of the right lung across the midline to the contralateral side. Displacement of the trachea to the left side was also noted.. On the 13th day of admission, further investigations revealed a raised ESR of 76mm/hr, negative induration following mantoux test (BCG vaccine was received at birth) and a chest ultrasound demonstrating irregularly thickened multiple cystic lesions within the right lung. During the course of admission, patient's condition improved and on the 16th day, all antibiotics were stopped. Due to financial constraints, the chest CT scan was delayed and ultimately done on the 25th day following admission. CT images showed ill-defined areas of increased attenuation predominantly within the right lower lobe and the ipsilateral middle lobe. Rounded, air-dense, cystic areas of varying sizes were found within the right upper and middle lobes consistent with a congenital cystic pulmonary anomaly, likely congenital pulmonary airway malformation involving the right bronchial tree and affecting the right upper and middle lobes. See Figure 3

Patient remained clinically stable with occasional cough and although the caregivers were adequately counseled on the need for surgical excision, they declined because of financial constraints.

DISCUSSION

These 3 reported cases have shown that congenital lung anomalies can be misdiagnosed in children in the absence of a high index of suspicion. There is thus the need to consider the possibility of congenital lung anomalies in children presenting either with recurrent Respiratory Tract Infections (RTIs), persistent symptoms despite appropriate management or unclear chest radiographic imaging. All our three patients were initially diagnosed and managed as pneumonia, until events ranging from the chest imaging picture, to non-response to treatment, necessitated further investigations. This pattern of presentation with recurrent respiratory infections and respiratory distress has been previously reported.[5-8,11.12] However, some patients may be asymptomatic only to be identified as incidental findings on radiological imaging examinations for other ailments[5-8,11.12]

The first infant reported had been treated severally for RTIs over a course of 3 months, before presenting and was a case of repeated missed diagnosis. The diagnoses of CPAM in all three were suggested following Chest radiograph and CT scan requested because of the peculiarity of their clinical features and chest radiograph. This is not peculiar to our center as such has been the experience in other centers in Nigeria and beyond. [6,13,14]Adeniyi et al in South Western Nigeria, diagnosed and managed an infant as a case of hydropneumothorax before the chest computed tomography scan revealed that the child had CPAM.[13]Disu et al in Lagos, South West Nigeria, initially presumed that a 13 day old neonate, who presented with increasing respiratory distress and an abnormal chest radiograph had pneumonia.[14] A revised diagnosis of CPAM was made after a CT scan. Even in developed countries there have been reports of a time lag of two or more years from onset of symptoms to postnatal diagnosis of CPAM. [15,16]It is important to note that the index patients had pulmonary symptoms requiring further imaging. Some other patients have been reported to be incidentally diagnosed following imaging for non-pulmonary symptoms. [10]

The diagnosis of pneumonia in these 3 infants and the

others in South West Nigeria is consistent with global practice for any child presenting with cough, fever and fast breathing. Most often, and following the directives of the World Health Organization for diagnosis of acute respiratory infections, infants and children presenting with cough, fever and fast breathing are diagnosed as having pneumonia and so treated.[17] They all presented at infancy with features suggestive of airway infection and failure to thrive which is typical of the clinical presentation at infancy.[5,16] However it is notable that some patients present even in adulthood as reported by previous authors with recurrent features of chest infection .[18]

The presentation and post natal diagnosis of CPAM in three children over the course of 3 years in one tertiary health facility in Nigeria, may suggest that such congenital lung anomalies, though rarely reported in developing countries like Nigeria, may be commoner than expected. This frequency may be comparable to what was reported in a tertiary centre in Scotland over a period of 14 years (1994-2004) where 26 children were diagnosed.[19]However all of them were diagnosed prenatally with no report of a post natal diagnosis. None of our patients was diagnosed in utero and apart from the second patient who had transient tachypnea shortly after delivery with occasional peripheral cyanosis, there was no indication at birth that these patients had a congenital lung anomaly. To date, there is no reported case of prenatal diagnosis of CPAM in Nigeria despite the fact that evidence has shown that most cases of CPAM are diagnosed in utero with the aid of prenatal ultrasound scan and MRI, with many regressing during pregnancy and only a small proportion presenting postnatally.[5,8,11,12,20] However, some studies have found only a 57% positive predictive value with prenatal diagnosis.[12]

This regional challenge with pre-natal diagnosis is expected as there is no policy on routine prenatal screening for pregnant women in Nigeria.[21] While carrying out an audit of routine ultrasonography done in a hospital in South West Nigeria, the number screened represented less than 5% of the yearly antenatal visits and no diagnosis of a congenital lung anomaly was made.[21] Though ultrasound is readily available in our environment, pregnant mothers are not routinely scanned. This service is usually paid for out of pocket.[21]

Using the Stocker classification of CPAM, our patients appear to fall into the groups with better prognosis (Type 1).[3] However a definite classification would have been made after histology of tissue sample, which could not be done as the parents of none of the patients could afford a surgical intervention. Some cases of CPAM may resolve spontaneously before birth while some are aborted spontaneously.[5,8,12] The gravest prenatal complication associated with this condition is fetal hydrops while postnatally, there are risks for malignant transformation arising from this malformation later in life if untreated .[1,2,5,6,12,15,22] Other complications include pulmonary hypoplasia from the effect of compression of large lesions and respiratory compromise.[1,2,6]None of the mothers of the 3 infants had adverse events in pregnancy and there was no report of features of hydrops in the infants. Most forms of CPAM usually occur in isolation as in the 3 infants who had no other identified congenital malformation.[2,5,6]However, some may present with gastrointestinal, cardiac, renal and skeletal abnormalities.[2,5,6]

Chest X rays are very useful in making diagnosis of CPAM but are not confirmatory. In Nigeria where a significant proportion live on less than one dollar a day, very few can afford a CT. The parents of the second patient could not afford to pay out of pocket for a CT scan and this caused a delay in diagnosis. The third patient did her scan after 3 weeks on admission also because of financial constraints. On Xrays, lesions appear as multilocular cysts with thin walls surrounded by normal lung parenchyma.[23] The chest xrays done on the index patients raised our suspicion of CPAM which was confirmed using CT scan. CT

scan imaging is usually diagnostic. [5,13,18] Further confirmation could be obtained with tissue biopsy for histology either via open or Video Assisted Thoracoscopic (VAT) techniques. Presently, there are no facilities for VAT in our center. Complications of CPAM could arise prenatally or postnatally with lung hypoplasia, fetal non-immune hydrops, pneumothorax, haemopneumothorax, haemoptysis, recurrent respiratory tract infections, and failure to thrive. None of our patients had prenatal complications.[5,8]

The main modality of treatment is surgical excision (lobectomy).[24] There are also various forms of prenatal treatments which have been tried with success especially with those who have developed fetal hydrops in utero including thoracocentesis and pleuro-amniotic shunt placement. [2,5,25]. The first patient was booked for elective lobectomy by the cardiothoracic surgeons but parents declined due to financial constraints and fear of the unknown. The parents of the second patient found it difficult to foot the bill of the index admission especially increased by the need for over 60 hours of intranasal oxygen. The parents of the third patient equally had financial constraints. None of these 3 reported cases eventually had required surgical intervention. Knowing the possibility of children with this condition to develop complications like malignant transformations and have recurrent respiratory tract infections before surgical intervention, the outlook in a developing country like ours with no sustainable or wide reaching health insurance scheme is bleak.

CONCLUSION

Congenital lung abnormalities such as CPAM do occur in our environment but may be missed due to paucity of diagnostic tools and cost implication for diagnosis. Even when diagnosed properly, poverty and ignorance on the part of the parents/care givers limit definitive care. Physicians need to have a high index for suspicion of congenital lung abnormalities in children, especially those who present with severe / recurrent pneumonia like illness. Parents and caregivers need support and financial empowerment to provide optimal care for such children.

Disclosures

The first case was used for a poster presentation at the 2018 ATS International Conference and published as an abstract in the American Journal of Respiratory and Critical Care Medicine 2020;201:A5620

Declarations

Availability of data and materials- Available on request

Competing interests- None

Funding- None

Consent for publication: Informed consent was obtained from the patients' parents for publication of these case reports and any accompanying images.

Acknowledgments: We are grateful to the parents of our patients for their cooperation.

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