

Variability of Presentation and Surgical Approach in Patients with Congenital Cystic Adenomatoid Malformation: Report of Two Cases

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Abstract

Congenital cystic adenomatoid malformation (CCAM) is a rare hypoplastic, dysplastic or hamartomatous disorder of lung. Two such cases were referred to our institute and managed surgically. The first case, a 4 month old male baby diagnosed antenatally with cystic lesions in the lung and suffered from respiratory tract infection postnatally. A computerised tomography scan (CT scan) was done to assess area of involvement in the lung. As apical and anterior basal segment (Segment 6 and 7) was found to be involved, segmentectomy was performed. The second patient, a 2 month old female baby, presented with features of pneumothorax with a normal perinatal history. CT scan revealed multiple cystic lesions of different sizes involving whole left lobe. Left lower lobectomy was performed. The authors emphasize on postnatal confirmation of CCAM by CT scan and lobectomy or segmentectomy as early as possible after 1 month of age guided by CT scan and thoracotomy finding even in asymptomatic patient.

Keywords: Congenital cystic adenomatoid malformation (CCAM); Congenital pulmonary airway malformation (CPAM); Congenital cystic lung disease; Congenital lobar emphysema; Ventilatory strategy

Introduction

Congenital cystic adenomatoid malformation (CCAM) is a rare dysplastic or hamartomatous disorder of lung characterised by overgrowth of terminal bronchioles with a reduction in the number of alveoli [1,2] resulting in formation of a multicystic mass of pulmonary tissue with an abnormal proliferation of the bronchial structure [3,4]. Incidence of CCAM varies between 1 in 25,000 to 1 in 35,000 pregnancies and it represents 25% of all congenital lung malformations and 95% of congenital lung lesions [5-8]. Advances in imaging modalities have enabled clinicians to diagnose CCAM in the antenatal period and intervene at the earliest if necessary [1,2,9-17]. There have been controversies among clinicians regarding optimal management and timing of intervention in CCAM patients [1,2,9,10]. Here the authors present successful surgical management of two cases of CCAM presenting with two different scenarios. The first case presented with antenatal ultrasound diagnosis and subsequent postnatal complications and the second patient presented with features of pneumothorax with a normal antenatal history.

Clinical Summary

Case 1

A 4 month old 6 kg male baby was referred to our institute with a history of antenatally diagnosed cystic lesions in the left lung. The mother attended antenatal clinic and routine ultrasound for anomaly scan in 18 weeks of pregnancy revealed multiple cystic spaces in left hemithorax with no abnormal mediastinal shift. The mother was 32 years old primigravida without any history of abnormal enlargement of abdomen or exaggerated nausea and vomiting in antenatal period. The pregnancy continued normally and was terminated by caesarian section at 39 week. No cardiopulmonary resuscitation of the baby was required after birth. After birth the baby was kept in neonatal intensive care unit for twenty two days for respiratory insufficiency and treated conservatively with moist oxygen. There was also a history of cough and nasal discharge with increased respiratory rate at the age of 2 months which was controlled with medications. A computerised tomography scan (CT scan) was done at the age of 3 months and digital scanogram in supine AP and lateral projection revealed ground glass

haziness in left lung with mediastinal shift to the right side. There was well defined, conglomerated, cystic (hyperlucent) lesion with lobulated margin in the left lower lobe along with left lower lobe collapse (Figure 1). The diagnosis of congenital cystic adenomatoid malformation was considered from the presentation and clinical and imaging finding. Hemogram and blood biochemistry was within normal limit. Patient was posted for definitive surgery. Chest was opened with left posterolateral thoracotomy with incision along 5th-6th intercostal space (Figure 2). The 5th rib was resected 4 cm for adequate exposure. The apical and anterior basal segment (Segment 6 and 7 left sides) was found to be involved without involvement of other segments. Segmentectomy was done and the unaffected lung expanded well with recruitment maneuver. Examination of the resected specimen revealed multiple small cysts involving the affected lobe with diameter <2 cm. Histopathological examination revealed multiple cysts lined by ciliated columnar epithelium consistent with type 2 CCAM. Patient was transferred to step down recovery on the third post operative day, to general ward on the 6th day and discharged after 10th day after removal of stitches. Postoperative course was uneventful.

Case 2

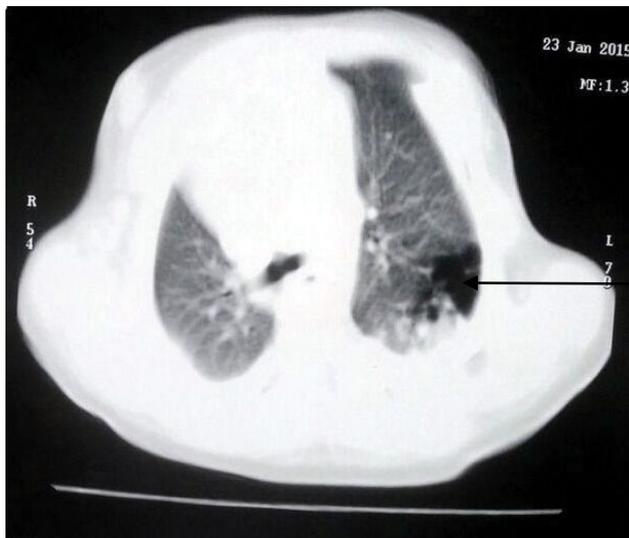
A 2 month old female baby was referred to this institute with history of respiratory distress with fever. Antenatal scan revealed no abnormality. Patient was admitted previously in a health centre and diagnosed as massive pneumothorax of left side from clinical examination and chest x-ray (Figure 3). A chest tube was inserted but no improvement occurred. After admission and stabilisation of the baby with conservative management, a CT scan of chest was done and it revealed pneumothorax, multiple cystic lesions of different sizes having

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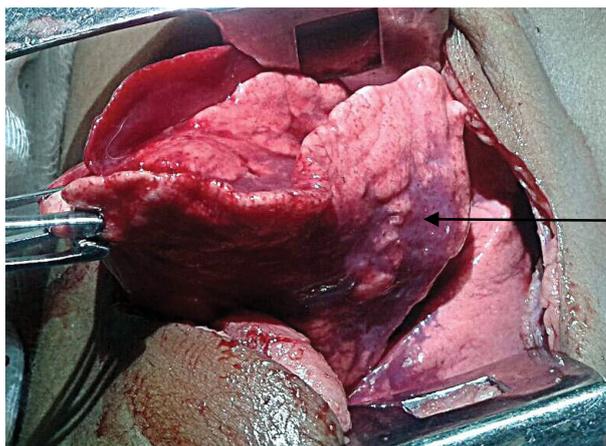
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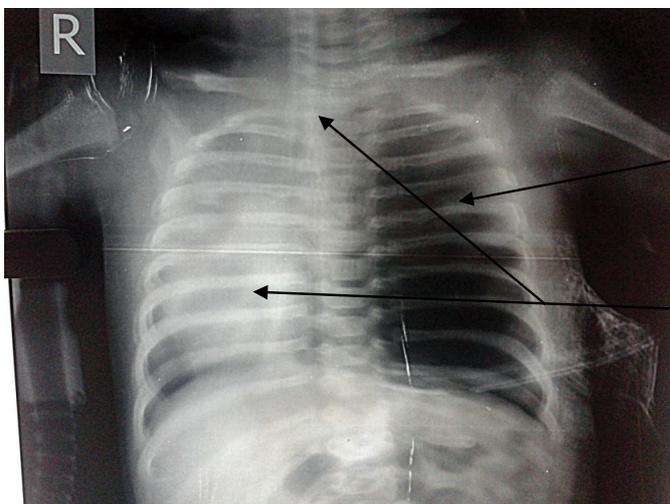
Affected segment

Figure 1: Well defined, conglomerated, cystic (hyperlucent) lesion with lobulated margin in the left lower lobe along with left lower lobe collapse (Patient 1).



Affected segments

Figure 2: Affected lung segments at thoracotomy (Patient 1).



Translucency at the affected side

Trachea and heart displaced at opposite side

Figure 3: Chest x-ray (Patient 2).

thin and thick wall, mediastinal shift towards right and compression collapse of right upper lobe. The trachea and the major bronchi and the great vessels of thorax appeared normal (Figure 4). The patient was diagnosed as CCAM and surgical exploration with removal of the affected lobe was planned. Left posterolateral thoracotomy was done and the whole left lower lobe was found to be involved (Figure 5). Left lower lobectomy was performed and the remaining lung expanded with recruitment manoeuvre. Patient was extubated on the second day. Examination of the resected specimen revealed multiple large cysts of varying size (Largest one being 4 × 4 cm) involving the left lower lobe. Postoperatively patient was managed with respiratory physiotherapy, antibiotics, analgesics and nebulised bronchodilators as required. Chest tube was removed after 3 days. The patient was discharged home after 6 days.

Discussion

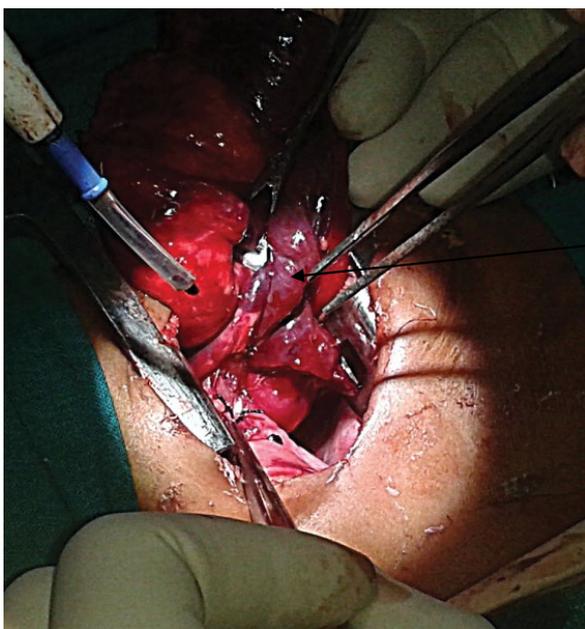
The definite pathophysiological mechanism behind CCAM is not yet understood. Many theories have been suggested including overgrowth [18], hyperplasia [19] and hamartoma [20] theory is currently considered among pathologists [1]. However it is distinct from other similar congenital cystic anomalies by several typical characteristics [21]. Firstly there is absent bronchial cartilage. Secondly bronchial tubular glands are absent. Thirdly, there is dominance of tall columnar mucinous epithelium. Fourthly there is overproduction of terminal bronchiolar structures without alveolar differentiation. Fifthly, there is proliferation of smooth muscle and elastic tissue in cyst walls. And lastly, there are normal arterial and venous connections. CCAM may be diagnosed antenatally or after birth. Large CCAM may be associated with life threatening complications such as fetal hydrops, cardiovascular dysfunction, lung hypoplasia etc and should be operated as early as possible. The pathogenesis behind pulmonary hypoplasia may involve compression of the developing lung tissue by enlarging CCAM mass [22,23] or bronchial atresia [24]. The probable mechanism of fetal hydrops is displacement of the mediastinum and compression of vena cava [23].

During the antenatal period polyhydramnios may be the warning sign of development of CCAM [25]. After birth children usually present around 48 months of age. Commonest presentations are respiratory distress in children under 6 months of age and recurrent pneumonia in older ones [26]. Other presenting feature include recurrent respiratory tract infection, cough, fever, haemoptysis or failure to thrive in the neonatal period or early childhood [27,28]. Physical signs include tachypnoea, signs of pneumothorax/air trapping including tracheal deviation, shifted heart sounds, and decreased air entry on the affected side; cyanosis, use of accessory muscles for respiration, grunting, etc. Common differential diagnoses include congenital diaphragmatic hernia, congenital pneumonia, haemothorax, pneumatocele and pneumothorax. [10] Factors indicating poor prognosis is described in Table 1 [29-33].

With advancement of imaging technique CCAM can be diagnosed

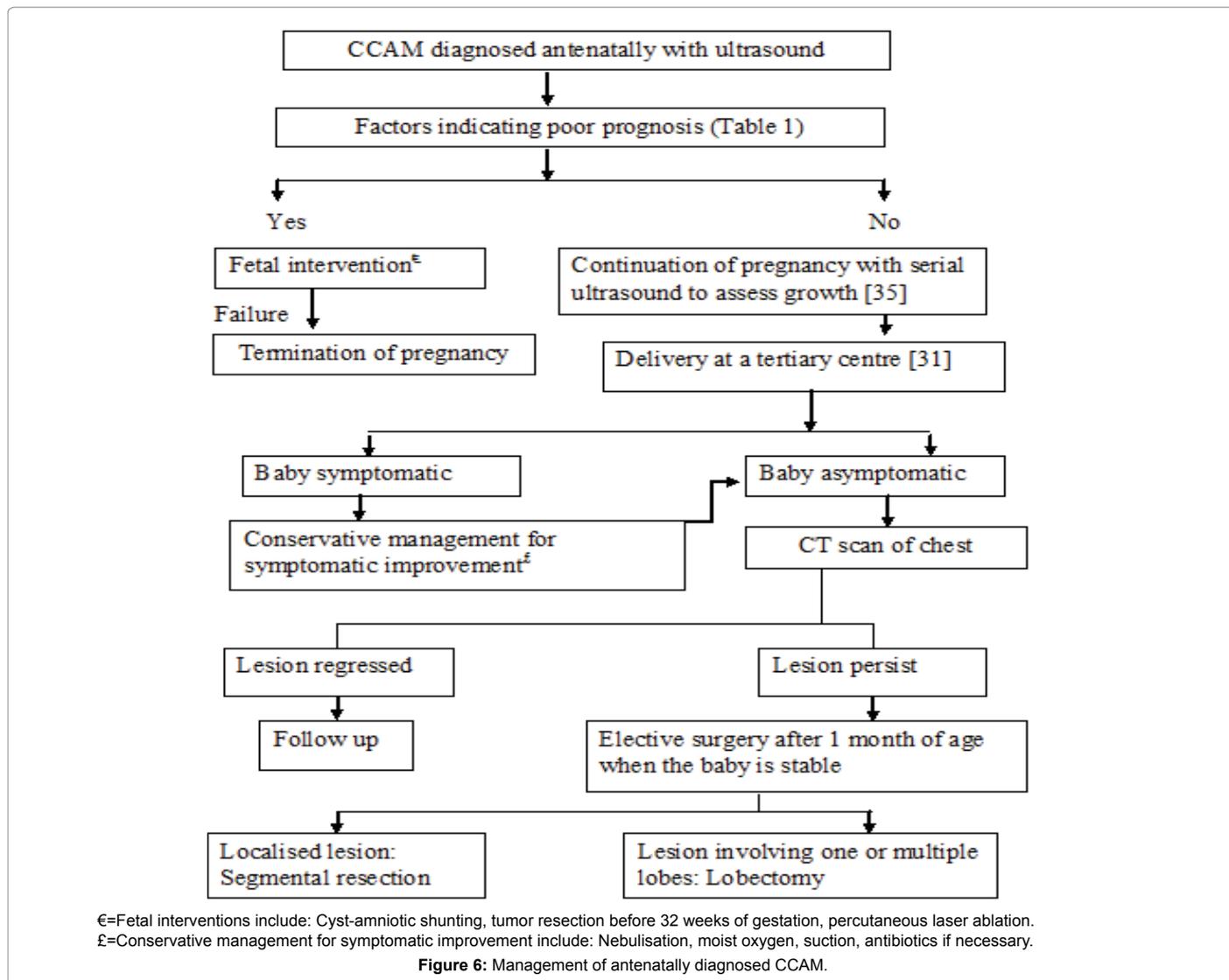


Figure 4: CT scan of chest revealed multiple cystic lesions of different sizes having thin and thick wall largest one measuring 5 × 2 cm (Patient 2).



Cysts in the affected lobe

Figure 5: CCAM lesion at thoracotomy.



- Hydrops fetalis,
- Ascites,
- Polyhydramnios,
- Bilateral lung involvement,
- Stocker's type III
- Lung-to-thorax transverse area ratio of less than 0.25
- Cystic adenomatoid malformation volume ratio-defined as the estimated volume of the CCAM divided by head circumference. A ratio >1.6, leads to a poorer prognosis.
- Severe illness in mother due to mirror syndrome

Table 1: Factors indicating poor prognosis in antenatal period [29,30-33,61].

antenatally with fair accuracy. Ultrasound is the antenatal imaging modality of choice for screening for CCAMs whilst magnetic resonance imaging is complimentary for morphological and volumetric evaluation of the fetal lung [1,2,8-16,34]. Prenatal regression and complete postnatal resolution may occur in the course of pregnancy in 76% cases [1,35]. In a patient with CCAM presence of fetal hydrops is the best predictor of death [1,25]. Computerised tomography scan is the best modality of accurately postnatal diagnosis and grading CCAM lesions [1,36].

There have been controversies regarding importance of prenatal

and postnatal diagnosis of CCAM. Although imaging modalities can fairly identify this pathology but the applicability and acceptability of antenatal diagnosis is questionable [29]. As in most of the cases the lesions regress spontaneously the patient may not require any therapy. In a small minority of foetuses large lesions may develop which may necessitate treatment in utero [29]. Postnatally the babies may develop respiratory symptoms or may remain asymptomatic for a long period. Prenatally diagnosed CCAM usually have a favourable outcome [37-39]. Termination of pregnancy may be required in patients with poor prognosis (Table 1). Both the mothers of the babies in this discussion did not have any stated signs in antenatal period.

Treatment of symptomatic CCAM is essentially surgical. Elective lobectomy is the most preferred surgical option [1,29,34,37]. There have been controversies regarding management of the asymptomatic cases [1]. Adzick [25] recommended resection of involved segment or lobectomy as early as possible after 1 month in asymptomatic cases to prevent development of future complications (Table 2) including malignancy and to promote compensatory lung growth. Similar views are shared by many other clinicians [40-42]. Collins and coworkers [43] emphasized on hazards of delayed diagnosis and surgery as potentially incomplete resections may increase the risk of complications and malignant transformation. Whereas some authors prefer to avoid surgery and follow up closely for development of complications. Their argument is the risk of development of malignancy is not well defined, cysts may reduce spontaneously and complications may be revealed by continuous follow up [1]. Butterworth and Blair [44] retrospectively reviewed cases of antenatally diagnosed CCAM over 7 years and opined in favor of an observational period before resection. Hsieh et al. [45] also opined in favor of conservative management.

During perioperative period, the CCAM mass may behave as a mediastinal mass as well as an emphysematous bulla and may bring about hemodynamic and respiratory complications. Administration of anesthetics cause reduction in tone of thoracic muscles abolishing the outward recoil tone and renders intrathoracic pressure to become positive during inspiration [46]. This may aggravate the compressive effect of the mass as well as produce de novo compression over heart or any vital intrathoracic structure. Overdistension of the cystic lesions during ventilation before the chest is opened may also cause similar effect [47,48]. To avoid overdistension multiple techniques have been described in literature and should be individualized according to the case (Table 3). In general gentle ventilation with low airway pressure (20-25 cm H₂O) keeping a close watch on the vital parameters is recommended. Nitrous oxide must be avoided as it diffuses rapidly into air filled closed spaces and enlarge its volume [49]. An arterial line is essential for monitoring beat to beat variation in blood pressure and blood gas analysis. A double lumen endobronchial tube may be used in older children and adults for differential lung ventilation. Severe desaturation may occur during induction or intubation and the anesthesiologist must be prepared to manage a difficult intubation

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| <ul style="list-style-type: none"> • Respiratory infections, • Hemorrhage • Sudden respiratory compromise • Pneumothorax • Malignancy: bronchoalveolar cancer and rhabdomyosarcoma more common |
|---|

Table 2: Complications of untreated CCAM [29,54,61,62].

scenario [50]. Awake intubation may be preferred technique in selected cases [51,52]. Analgesia is crucial in thoracotomy patients and multimodal analgesia technique is preferred. The authors used a caudal epidural catheter for postoperative pain management along with paracetamol suppositories for the initial 48 h.

The outcome of surgery differs with the characteristics of the lesion and clinical presentation. Aziz et al. [46] performed a retrospective review of hospital records between 1996 and 2002 in a tertiary care pediatric referral centre and found similar postoperative morbidity in complicated and uncomplicated CCAM. Elective surgery is usually well tolerated, the risk of infection is lower, and compensatory lung growth with normal long-term respiratory function has been observed. Clinicians are reconsidering this approach in the light of the spontaneous improvement and possible resolution that occurs over months to years with many of these lesions, thinking about the opportunity to take a more conservative approach in many minimally symptomatic or asymptomatic infants in the early months of life [1,53-59].

There have also been controversies regarding the age of surgery in asymptomatic patients. Early surgery may be associated with adequate growth and expansion of the remaining lung to restore the total lung volume and satisfactory pulmonary function test [26]. Adzick recommended surgery after the age of 1 month [32]. Some authors advocated surgery between 6 and 12 months of age because anesthetic and surgical risks decrease within the first months of life [29]. Kotecha et al. [34] recommended surgery within first year of life. Calvert and Lakhoo [60] suggested resection between 3 and 6 months of age of all CCAMs diagnosed prenatally to avoid infection risk which is much more common if resected after 6 months. The authors are in favor of surgery even in asymptomatic cases as early as possible after 1 month. The cases in discussion were operated at 3 and 4 months and they tolerated the procedure well [61,62].

The extent of surgery is also important. Most authors are in favor of lobectomy [1,25]. In selected cases segmental resection was also found to be associated with favorable outcome. The authors recommend decision of extent of surgical excision should be guided by the postnatal CT scan and examination of the affected segment during surgery. The first case in this discussion was treated with segmental resection as involvement was limited the specific segments whereas in the second case lobectomy was required to remove the diseased area.

In the view of currently available evidences in literature, the authors recommend an integrated approach to antenatally diagnosed CCAM which is depicted in Figure 6.

Recommended by	Strategy	Physiological basis
Payne et al. [32], Cote [33]	Manual ventilation maintaining the airway pressure at 20-25 cm H ₂ O before thoracotomy, keeping an eye on the vital signs	Avoid over distension by reducing ventilator pressure
Mandelbaum et al. [34] Loewy et al. [35] Kumaghai et al. [31] Hugh et al. [30]	Selective endobronchial intubation of the healthy lung till the lobectomy/ pneumonectomy is performed, after which the endotracheal tube is withdrawn in the mainstem bronchus	Avoid ventilation of the affected lung
Goto et al. [36]	High frequency jet ventilation	Low airway pressure, Minimal change in airway pressure
Raghavendra et al. [37]	Thoracic epidural catheter via the caudal insertion site, adequate analgesia and retaining spontaneous ventilation until thoracotomy	Avoid intubation until thoracotomy
Phillipos et al. [38]	A flexible ultra-thin bronchoscope into the bronchus of affected side	Relieve obstruction
Campbell et al. [39]	Continuous Positive Airway Pressure	

Table 3: Ventilatory strategy to avoid over distension in cystic lesions of lung.

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