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Original article

Congenital cystic adenomatoid malformation: the experience of a level III Neonatal Intensive Care Unit

Nuno Medeiros[1], Gustavo Rocha[2], Tiago Henriques-Coelho[1,3], Carla Ramalho[1,4,5], Hercília Guimarães[1,2]

[1]Faculty of Medicine, Porto University, Porto, Portugal
[2]Neonatal Intensive Care Unit, Centro Hospitalar de São João, Porto, Portugal
[3]Department of Pediatric Surgery, Centro Hospitalar de São João, Porto, Portugal
[4]Department of Obstetrics and Gynecology, Centro Hospitalar de São João, Porto, Portugal
[5]Institute of Health Research and Development, Porto University, Porto, Portugal

Abstract

Background: Congenital cystic adenomatoid malformation (CCAM) is a rare disease. Because few cases per center are reported each year, there is still uncertainty concerning pathophysiology, natural history of the disease and the best treatment approach.

The aim of this paper is to report the experience of a level III Neonatal Intensive Care Unit (NICU) in CCAM cases.

Methods: We evaluated all cases diagnosed with CCAM admitted to our hospital between January 2000 and December 2014.

Results: Twenty-four neonates with prenatal diagnosis of CCAM were identified. The majority of lesions (75%) had microcystic features. Thirteen newborns were admitted to NICU for observation and evaluation. Half of the prenatally diagnosed lesions were confirmed cases of CCAM. The remaining corresponded to 5 cases (20.8%) of *in utero* spontaneous regression of the lesion, 5 (20.8%) bronchopulmonary sequestration, 1 (4.2%) bronchial atresia and 1 (4.2%) pleuro-pericardial cyst. Two (8.3%) neonates became symptomatic during the neonatal period and surgery was performed in the first week of life. Ten cases remained asymptomatic. Six of these cases (40%) underwent surgery (thoracotomy in 3 cases and thoracoscopy in 3 cases). Respiratory morbidity was reported in 1 symptomatic case and in 1 asymptomatic case that was managed conservatively. No mortality was reported. Histological examination showed definitive features of CCAM (Stocker classification: type I = 1; type II = 4; type III = 2).

Conclusion: Management of CCAM in a NICU does not seem necessary in asymptomatic newborns. Concordance of ante-natal and post-natal findings is very variable. Thoracoscopy is a less invasive surgical procedure in the management of these patients. The outcome is good in most patients.

Keywords

Congenital cystic adenomatoid malformation of lung, neonatal intensive care unit, thoracoscopy.

Corresponding author

Nuno Miguel Medeiros Oliveira, Faculty of Medicine, Porto University, Alameda Prof. Hernâni Monteiro, 4200-319 Porto, Portugal; email: nuno_mmoliveira@hotmail.com.

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Introduction

Congenital cystic adenomatoid malformation (CCAM), also known as congenital pulmonary airway malformation (CPAM), is the most common congenital lung malformation. However, it is still a rare disease with an incidence between 1/25,000 and 1/35,000 cases per year [1]. It represents an embryologic anomaly of the development of the terminal bronchi, which proliferate and enlarge, leading to cyst formation and absence of alveoli. They are still connected to the tracheobronchial tree and have normal pulmonary blood supply [2]. The pathogenesis of CCAM is still poorly understood, and theories include a primary foregut development anomaly or dysplastic changes secondary to airway obstructions.

In advanced countries with an efficient health care system, CCAM is detected during obstetric ultrasound (US) in the vast majority of cases (99%). Whenever no prenatal care is given, CCAM may be found after delivery and represents a diagnostic challenge. Some lesions detected prenatally seem to have completely disappeared at term, but further postnatal studies may show their presence, playing an important role in the diagnosis of CCAM [1]. Although magnetic resonance imaging (MRI) exposes the newborn to less radiation, it would imply sedation; hence, computerized tomography (CT) is the most used imaging technique in the postnatal period. X-ray has limited value as it only identifies 61-67% of the cases [3]. CT seems to have a sensibility of 100% [4]. There is little evidence suggesting that lesions that are present postnatally may undergo regression [5].

The most used classification for CCAM is still the Stocker classification that subdivides the findings in grades from 0 to IV [6]. Recently, the Adzick classification is acquiring its relevance. It divides CCAM into two categories, macrocystic and microcystic, on the basis of a 5-cm cut-off [5].

This study aimed to evaluate the experience of our level III Neonatal Intensive Care Unit (NICU) with CCAM and to compare it to literature data, in order to improve our practice in prenatal diagnosis and clinical management of newborns.

Population study and methods

We conducted an observational retrospective study. Cases of CCAM suspected on prenatal US were selected from the newborn nursery and the Neonatal Unit at "Centro Hospitalar São João", Porto, between January 2000 and December 2014. There were 42,771 births registered at this hospital in this period. From these reports, the description of the imaging examinations performed on the newborn in the first days of life were analyzed to assess if they corresponded to true CCAM cases or if other diagnoses were established afterwards. Newborns were followed until 2014 to ensure that they all were at least 12 months old when data were collected and analyzed.

The following data was collected: maternal age, gestational age at diagnosis, gestational age at birth, gender, birthweight, multiple pregnancy, affected lung, need for NICU admission, final diagnosis, Stocker classification of the lesion (type 0 – small solid lungs; type I – cysts up to 10 cm lined by pseudostratified ciliated cells; type II – sponge-like multiple small cysts (< 2 cm); type III – excess of bronchiolar structures separated by small air spaces with cuboidal lining, microscopic adenomatoid cysts; type IV – cysts up to 10 cm lined by flattened epithelium), associated malformations, symptoms at birth, timing of surgery, type of surgery, morbidity after surgery and overall mortality.

The study was approved by the Ethics Committee of our center.

Data was collected by the same researcher and recorded in a digital database (Statistical Package for the Social Sciences – SPSS® 22). Given the size of the sample, it was only possible to conduct a descriptive analysis.

Results

Antenatal US evaluation suggested the existence of 24 cases of CCAM (**Fig. 1**). In 3 of the cases, the existence of a congenital lung malformation suggested by US was also confirmed by prenatal MRI. Antenatal karyotyping was not carried out in any of the patients. Prenatal diagnosis was performed between 19 and 27 weeks (with a median of 21 weeks).

Tab. 1 summarizes data regarding mothers and the 24 newborns in the perinatal period. All pregnancies resulted in live births at a median gestation time of 39 weeks (35-41 weeks) and with a median birthweight of 3,235 g (2,275-4,160 g). Twelve (50%) cases were males. Two of the 3 cases born before completing 37 weeks of gestation corresponded to twin pregnancies.

Data on prenatal imaging features, NICU admission and final diagnosis are reported in **Tab. 2**. Fourteen (58.3%) newborns were admitted to the NICU for observation and evaluation, while

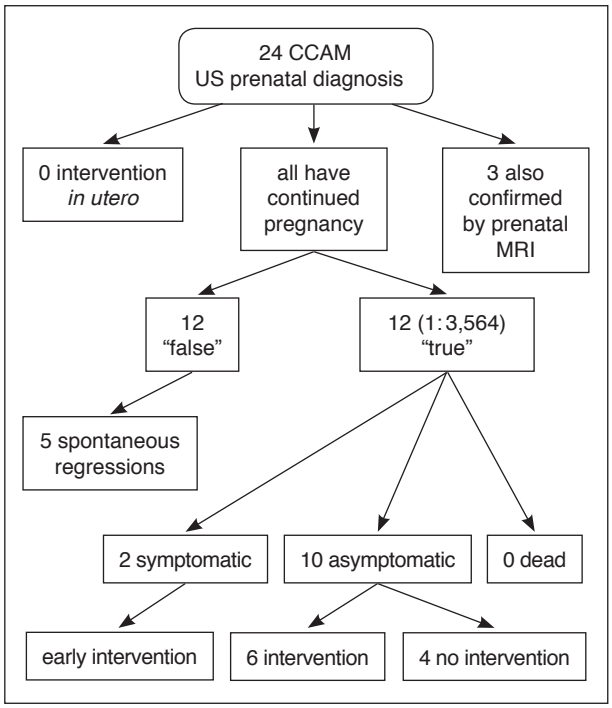

Figure 1. Flow chart of the population of the study.
CCAM: congenital cystic adenomatoid malformation; MRI: magnetic resonance imaging.

Table 1. Perinatal data of congenital cystic adenomatoid malformation (CCAM) cases detected by ultrasound (US) during pregnancy.

Case no.	Year	Maternal age	Gestational age at pre-natal diagnosis (weeks)	Gender	Gestational age at birth (weeks)	Weight at birth (g)
1	2001	27	Unknown	F	39	3,850
2	2004	19	20	M	40	4,030
3	2005	29	21	F	37	3,975
4	2007	34	25	F	40	3,230
5	2008	22	22	F	38	3,470
6	2008	32	27	F	39	3,100
7	2009	38	23	M	36	3,180
8	2009	33	21	F	39	3,140
9	2009	39	21	M	39	3,420
10	2009	28	22	F	39	4,000
11	2011	36	22	M	39	2,990
12	2011	28	21	M	41	3,620
13	2011	30	20	F	41	2,600
14	2011	29	21	M	39	3,120
15	2012	32	21	M	39	3,240
16	2012	36	24	M	36 (1st twin)	2,300
17	2013	39	19	M	39	2,990
18	2013	30	21	M	39	3,650
19	2013	37	19	F	35 (1st twin)	2,275
20	2013	35	21	F	40	3,630
21	2013	41	21	M	38	2,865
22	2013	33	22	F	39	3,445
23	2014	31	21	F	39	3,230
24	2014	31	22	M	38	4,160

Table 2. Pre-natal and post-natal diagnosis.

Case no.	Affected lung	US diagnosis	Prenatal MRI	Admitted to NICU	Final diagnosis[a]
1	Right	Small cyst	No	Yes	CCAM
2	Left	Nonspecific	No	Yes	CCAM
3	Left	Small cyst	No	Yes	CCAM
4	Bilateral	Nonspecific	No	Yes	CCAM
5	Bilateral	Large cyst	No	Yes	Pleural-pericardial cyst
6	Left	Small cyst	No	No	Regression
7	Right	Large cyst	No	Yes	CCAM
8	Left	Small cyst	Yes	No	CCAM
9	Left	Large cyst	No	Yes	CCAM
10	Right	Nonspecific	No	No	CCAM
11	Right	Small cyst	Yes	Yes	BPS
12	Left	Small cyst	No	Yes	Complex lesion (CCAM/BPS)
13	Right	Small cyst	No	Yes	CCAM
14	Right	Small cyst	No	No	CCAM
15	Right	Small cyst	No	No	BPS
16	Left	Small cyst	No	No	Regression
17	Right	Small cyst	No	No	BPS
18	Right	Small cyst	No	No	Regression
19	Left	Small cyst	No	No	Regression
20	Right	Small cyst	No	Yes	BPS
21	Left	Small cyst	No	Yes	Bronchial atresia
22	Left	Small cyst	No	Yes	BPS
23	Right	Small cyst	No	No	CCAM
24	Left	Small cyst	Yes	Yes	Regression

US: ultrasound; MRI: magnetic resonance imaging; NICU: neonatal intensive care unit; CCAM: congenital cystic adenomatoid malfomation; BPS: bronchopulmonary sequestration.
[a]Post-natal X-ray and/or CT scan were performed. Only in 2 cases a CT scan was not performed during the neonatal period. In these cases (cases no. 6 and 19), only an X-ray was performed

10 (41.7%) stayed in the nursery for healthy newborns in the Obstetrics department.

According to imaging evaluation, the lesions affected only the right lung in 11 newborns, only the left lung in other 11 newborns, and were bilateral in 2 cases. US features denoted small cyst lesions in 18 (75%) cases, large cyst lesions in 3 (12.5%) cases and nonspecific features in 3 (12.5%) cases.

Two (8.3%) newborns were symptomatic at birth (respiratory distress and mediastinal shift) and the other 22 (91.7%) were asymptomatic at birth. Both symptomatic children had a confirmed CCAM lesion at pathological diagnosis, type I and type III respectively.

When post-natal X-ray and/or CT scan were performed, the final diagnosis corresponded to 12 (50%) cases of CCAM (1 of which was a complex lesion), 5 (20.8%) cases of bronchopulmonary sequestration (BPS), 1 (4.2%) case of bronchial atresia and 1 (4.2%) case of a pleural-pericardial cyst. In 5 (20.8%) cases, the cystic lesion identified in the antenatal US was not found in post-natal studies.

There were 12 confirmed cases of CCAM in this 15-year period study. The registered incidence in this specialized center was 1:3,564.

The location of the lesion was confirmed in the majority of cases. In 1 of the bilateral cases (case no. 5), the lesion was proved to be a pleural-pericardial cyst in the mediastinum. In 1 patient (case no. 1), that had only a described right lung lesion, other cystic lesions were also found in the lingula.

Tab. 3 presents post-natal data and outcomes of all the postnatally confirmed CCAM cases.

Surgery (thoracotomy) was performed in the first week of life in both symptomatic newborns (cases no. 7 and 9). Four of the remaining CCAM cases did not undergo surgery and the other 6 underwent surgery between 9 months and 3 years of age. They corresponded to 3 thoracotomies (cases no. 1, 2 and 3) and 3 thoracoscopies (cases no. 12, 13 and 23). Case no. 3 underwent partial lobectomy,

Table 3. Post-natal management of confirmed congenital cystic adenomatoid malformation (CCAM) cases.

Case no.	Symptoms at birth	Timing of surgery	Type of surgery	CCAM type[a]	Morbidity after surgery
1	No	9 months	Thoracotomy	III	No
2	No	2 years	Thoracotomy	II	No
3	No	18 months	Thoracotomy	II	No
4	No	No surgery	-	-	Multiple respiratory infections
7	Yes	1st week	Thoracotomy	III	Multiple respiratory infections
8	No	No surgery	-	-	No
9	Yes	1st week	Thoracotomy	I	No
10	No	No surgery	-	-	No
12	No	1 year	Thoracoscopy	II	No
13	No	3 years	Thoracoscopy	II	No
14	No	No surgery	-	-	No
23	No	1 year	Thoracoscopy	-	No

[a]Histological diagnosis, Stocker classification.
CCAM: congenital cystic adenomatoid malformation.

while the others underwent total lobectomy. After the anatomopathological examination of the lesions and using the Stocker classification, we diagnosed 1 case of type I CCAM, 4 cases of type II and 2 cases of type III. No cases of type 0 or type IV CCAM were reported.

The rate of complications was low, with frequent respiratory infections being reported in 1 of the cases surgically managed and in 1 of the cases managed with a non-invasive approach. Mortality was not recorded up to the date of conclusion of this study.

Discussion

Our data shows that the incidence of CCAM at our centre is 1:3,564, a high rate compared to the literature [1]. However, "Centro Hospitalar São João" is a referral center for surgical management of congenital lung malformations and for this reason this data is not reflective of the general population. Some cases with a prenatal diagnosis were in fact referred to this center.

According to literature, male fetuses are traditionally more likely to be affected by CCAM [2]. Our results do not support this statement, as there were more female newborns with confirmed CCAM than males in our series.

Prenatal diagnosis is usually performed between 20-22 weeks [7], similar to what we found in this study.

Some cases of antenatally suspected CCAM were after confirmed to be BPS or complex lesions (with features of both CCAM and BPS). Although prenatal US can give a clue about the provenience of the vascularization of the lesion, this is better evaluated postnatally. This fact supports the theory that these two conditions may represent different aspects of the same disease. Clements and Warner suggested that the major bronchopulmonary malformations are all lesions of the same disease spectrum, and that the dominant features depend on the timing of the interruption of the development, as the primitive systemic vascularization slowly regresses and the pulmonary supply becomes dominant [8].

MRI was used prenatally to confirm the diagnosis suggested by US in 3 of the cases (case no. 8, 11 and 24), including a case that was afterwards diagnosed as BPS. This finding is supported by another study claiming that no method can give a complete characterization of the lesion *in utero*, and that the additional information given by MRI may not necessarily imply changes in management [9]. Most sonographic features do not strongly correlate with neonatal outcome, however, the maximum and the final congenital pulmonary airway malformation volume ratio (CVR) seems to be an exception. The most sensitive cut-off value is yet to be defined [10].

In this series, the great majority of lesions had a small cyst appearance. There was agreement between antenatal and postnatal diagnosis in most of the CCAM cases, with the only type I lesion being correctly interpreted prenatally as large cyst. Another lesion with large cyst appearance at US was found to be a pleural-pericardial cyst. Type II and III of Stocker correspond to small cystic lesions

and only 1 case was interpreted as large cystic in prenatal findings. Interestingly, all the lesions that regressed *in utero* had small cystic features in US findings. This may indicate that regression of lesions may be associated with specific sonographic features. A larger case series might confirm or refute this hypothesis.

Many studies also report regression of lesions detected during pregnancy, as it occurred in 5 newborns in our series. However, although some lesions may appear to regress on serial US, they are usually detected by postnatal CT [11]. In our cases, 5 of the CCAM cases that were diagnosed prenatally were not visualized in postnatal studies. Only in 2 cases a CT scan was not performed during the neonatal period. In these cases (cases no. 6 and 19), only an X-ray was performed. CT scan has a sensitivity of ~100% but X-ray has a sensitivity of only 61% in detecting these lesions. We cannot conclude whether or not these were true CCAM cases, as imagiological regression is described in literature, and because in these 2 particular cases the study was not complete. CT scan is the best exam to confirm or exclude the pathology and should be performed before discharge.

The reported incidence of type I lesions was extremely low, with only 1 confirmed case at pathological examination. This finding is not supported by other studies. The most prevalent in our series was type II, which is reportedly the second most common type, followed by type III [6]. Type II and type III lesions are associated with a worse prognosis and higher incidence of morbidity, as the lesion usually grows more quickly. Surprisingly, most of our cases had a benign course, despite the small cyst appearance. On the other hand, case no. 9 presented with symptoms at birth, despite having a large cyst-type lesion, which is associated with a better prognosis. However, no post-surgical morbidity was reported.

As we verified in this study, less newborns with CCAM are admitted to a NICU in recent years. This occurs probably because of the increasing evidence that asymptomatic children can in most cases benefit from a perfectly normal life without any kind of special care [12]. The risk of future complications seems to be fairly acceptable, with some studies suggesting an incidence of 10%. This would mean that far too many children would be exposed to surgical risk unnecessarily if there were no risk of malignancy and serious infections [12]. In our asymptomatic cases in whom surgery was not performed, case no. 4 had a history of multiple respiratory infections during childhood. However, this may have been associated to a presence of bilateral lesions and not specifically to the management strategy.

Resection is currently undertaken in approximately two-third of pediatric surgical centers worldwide, regardless of lesion size or presence of symptoms [13]. When the newborn presents symptoms such as respiratory distress, or imaging signs like mediastinal shift, polyhidramnios or hydropsy, prompt intervention by a pediatric surgeon is advised to excise the lesion [14]. Case no. 7 underwent surgery but also presented with multiple respiratory infections. This child, however, had symptoms of respiratory distress in the neonatal period.

On the other hand, specialists advise that surgery is performed earlier than 6 months of age in asymptomatic cases, as it can reduce complications and maximize the compensatory growth of other lobes [15]. However, in a study performed with the follow-up at 2 years of age, no difference was shown in long-term pulmonary function [16]. Postnatal lung function also seems to be a parameter that can have some relevance in the non-invasive versus surgical dichotomy, as some CCAM can cause restrictive ventilation disorders [17]. In our asymptomatic cases, surgery was always performed after 6 months of age and we recorded no declared post-operative complications nor morbidity during follow-up in these patients. These cases illustrate that there are pros and cons in each treatment approach. In a study conducted by our department from 1999 to 2007, similar conclusions were reached, highlighting the good prognosis of this disease and the controversy in its treatment [18].

In recent years, thoracotomy has been sometimes replaced by minimally invasive thoracoscopy [19, 20]. The choice between these two methods is made on the basis of the experience of the surgical team and the localization and size of the lesion [21]. In our center, thoracoscopic lobectomy has been performed since 2011 for the management of CCAM and none of the 3 cases reported surgical-associated morbidity.

In all cases, except for 1 (case no. 3), lobectomy was performed. This is in agreement with several studies that demonstrated that pre-operative image techniques have a low sensitivity in detecting the limit between lesion and normal parenchyma. Although CT scan has a great sensitivity for detecting cystic lesions, it has a poor sensitivity to show the presence of lesions distant from the main cyst within the same lobe [22]. The use of atypical

pulmonary resections can be advocated in cases of multifocal CCAM lesions [7]. In case no. 3, a lobectomy was scheduled but the surgeon made an intra-surgical decision of an atypical resection of only some segments. As of today, there has been no recurrence of the lesion.

Diseases with this reduced incidence are more difficult to study because of the small number of patients. Small studies like this one contribute to a better knowledge of the disease, but they also demonstrate the importance of conducting larger studies with larger databases, to reach statically significant conclusions about the best features to evaluate in the antenatal and postnatal period, as well as the best treatment approach and follow-up strategy.

Conclusion

We can conclude that: 1) the natural history of prenatally diagnosed CCAM is highly variable; 2) the admission of patients to the NICU is becoming less frequent in recent years; 3) *in utero* sonographic features cannot completely characterize CCAM lesions; 4) regression of lesions is common and it may be more associated to specific sonographic features; 5) all cases of CCAM must be confirmed postnatally by CT scan; 6) surgical approach of symptomatic newborns is becoming less invasive, and excellent results can be obtained if performed by experienced surgeons; 7) strict timing of surgery does not seem essential.

Our main messages are that management in a NICU does not seem necessary in asymptomatic newborn, agreement of antenatal and postnatal findings is very variable and thoracoscopy is a less invasive surgical procedure in the management of these patients. We observed that most of the patients who are accurately managed, i.e. those with prenatal diagnosis, have a good outcome.

Declaration of interest

The Authors declare that there is no conflict of interest.

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