

Received: 2017.07.11
Accepted: 2017.07.14
Published: 2017.07.29

Computed Tomography (CT) Findings of Pulmonary Hydatid Cysts in Children and the Factors Related to Cyst Rupture

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
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Source of support: Departmental sources

Background: The lungs are the most common site of hydatid cysts in children. Rupture is the main complication of cysts causing morbidity and mortality. In this study we aimed to comprehensively describe the CT signs for cysts and analyze the relationship of cyst ruptures to the diameter and location of cyst.





Material/Methods: A total of 145 cysts from 102 patients, aged 17 years or younger, who underwent a multi-detector computed tomography (MDCT) evaluation and had surgically proven pulmonary hydatid disease were included retrospectively. The CT images were analyzed for radiologic findings and signs of cyst rupture.

Results: The cysts had a mean diameter of 5.45 ± 3.03 cm. Most of the patients had a solitary cyst (70.6%). The most common lobes involved were the lower lobes (58.6%). Peripherally located cysts were more common than central cysts. The overall number of ruptured cysts was 69 (47.5%). The most common sign was the waterlily sign with a prevalence of 24.6%. After exclusion of cysts of 1 cm, the mean cyst diameter was 6.23 ± 2.83 cm in the unruptured group and 5.02 ± 2.80 cm in the complete ruptured group ($p=0.020$). The mean cyst diameter was 6.38 ± 3.11 cm for centrally located cysts, and 4.31 ± 2.37 cm for peripherally located cysts ($p<0.0001$).

Conclusions: Pulmonary hydatid cysts commonly presents as solitary cysts with a predilection for lower lobes and the peripheral regions of the lung. Peripherally located cysts are more common but smaller than centrally located cysts. Radiologists should also be aware of atypical imaging findings of cyst rupture, and radiologic signs should be explored during radiologic evaluation.

MeSH Keywords: **Child • Echinococcosis, Pulmonary • Rupture • Tomography Scanners, X-Ray Computed**

Full-text PDF: <https://www.medscimonit.com/abstract/index/idArt/906163>

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Background

Hydatid disease is a worldwide parasitic disease caused by the larval stage of *Echinococcus granulosus*. The disease is endemic to central Asia, northern and eastern Africa, Australia, South America, and Mediterranean regions such as eastern and southeastern of Turkey [1].

In humans, hydatid disease involves the liver in approximately 60–75% of cases, the lung in 15–30% of cases, and less commonly involves other anatomic locations. The lungs are the second most frequent site of hematogenous spread in adults and the most common site in children [2–6].

The lungs facilitate cyst growth due to the lung's negative pressure and compressible nature [6], which may be the reason for the high prevalence of the disease in the lungs and easy growth of cysts in children. Most cysts are acquired in childhood, remain asymptomatic for a long period of time, and are later diagnosed incidentally on chest radiography. Pulmonary hydatid cysts are multiple in 30% of cases, bilateral in 20% of cases, and mainly located in the lower lobes in 55–70% of cases [2–9]. Calcification in pulmonary cysts is rare (0.7% of cases) [4].

Rupture is the main complication of cysts and may be related to the presence of symptoms, and to morbidity and mortality [8,9]. The rupture rate of hydatid cysts is quite high in children [10]. Rupture of a cyst into the pleural space or bronchus may even cause anaphylactic reactions [11]. Furthermore, obstruction of the tracheobronchial tree with cystic membrane can lead to suffocation [12].

Although the diameter of the cyst has been proposed as a risk factor for cyst perforation [13] some authors [14] have reported an enlargement in the size of the cyst as a negative risk factor for perforation. On the other hand, others authors [9,15] have reported no relationship between the size of the cyst and the risk of perforation. Recently, the lobe location of the cyst has been reported as a risk factor of perforation [9]. Many researchers have included both children and adults in their analysis, which may account for inconclusive results. The relationship between location (central versus peripheral), lobar distribution, size of the cyst, and rupture are still debated in the literature.

Herein, we aimed to comprehensively describe CT signs and findings of hydatid disease in children, and analyze the relationship of cyst diameter, lobar distribution, central versus peripherally location, and rupture of cysts.

Material and Methods

Patients

The study was designed as a descriptive retrospective study. We reviewed all patients diagnosed with pulmonary hydatid cyst disease admitted at Dicle University Hospital between 2007 and 2017. A total of 102 patients, age 17 years or younger, who underwent a multi-detector computed tomography (MDCT) evaluation and had surgically proven pulmonary cyst hydatid disease were included. Patient demographics, including age and gender, were obtained from the medical records of the hospital. The study was approved by local ethic committee (Number of 2017/139).

Imaging

All of the imaging findings of 102 patients who had undergone evaluation with 16-MDCT Toshiba Aquilion (Toshiba Corporation, Medical Systems Company), 64-MDCT scanner (Brilliance CT scanner, Philips Healthcare), and 256 MDCT (Siemens SOMATOM Definition Flash) were analyzed. The images were analyzed regarding cyst number, diameter, location, airway compression, presence of extrapulmonary cysts, calcification, and signs of cyst rupture.

All images were obtained from picture archiving and communication system (PACS) and evaluated at the work station. We used sagittal and coronal reformatted images in addition to axial images.

Definitions

The contained rupture was defined as detachment of the pericyst from the endocyst with the radiologic signs shown in Table 1. A complete rupture was defined in the presence of signs of rupture communicating with the bronchus. Signs for contained and complete cyst rupture were modified from previous studies [3–6]. Air between the pericyst and the endocyst created by bronchial erosion was defined as the “crescent sign”. Air crescent at the posterior aspect of a lesion caused by dissection through the posterior side of membranes was defined as “inverse crescent sign”. Air bubble sign was defined as small intracystic air focuses located at the periphery of cyst. Presence of air fluid level within the endocyst and the air crescent among the pericyst and endocyst was described as “cumbo sign” and collapsed membranes seen within the cyst after waste of cyst fluid as “whirl sign”. The wrinkled endocyst floating on fluid caused by complete collapse of cyst was named as “waterlily sign”. The wrinkled endocyst settled in lower parts of the cyst cavity was labeled as “mass within the cavity” and the empty and air filled cyst seen after appears expectoration of cyst content as “dry cyst sign”. We

Table 1. CT signs of rupture of hydatid cysts.

Signs of contained rupture	Signs of cyst complete rupture
Air crescent sign	Cumbo sign
Inverse crescent sign	Whirl sign
Air bubble sign	Waterlily sign
	Mass within a cavity sign
	Dry cyst sign

defined pulmonary hydatid cysts with a diameter of 10 cm or longer as “giant”.

Statistics

Continuous variables were expressed as mean \pm standard deviation and categorical variables as percentages (%). Categorical variables were compared using the chi squared test. Fisher exact test was used for statistical significance when a value less than 5 was present in 2x2 tables. Student’s *t*-test was applied to compare two groups regarding continuous variables. A *p* value <0.05 was considered significant. We compared giant versus non-giant groups of cysts and perforated versus non-perforated cysts with regards to demographic and radiological findings.

Results

A total of 102 pediatric cases (63 male, 39 female) with a mean age of 9.6 ± 3.9 years (range 0–17 years) were included. MDCT findings of 145 cysts from 102 patients were analyzed.

The cysts had a mean diameter of 5.45 ± 3.03 cm (range 1–13 cm). Most of the patients had a solitary cyst (70.6%); 24.5% of patients had two cysts, and only 4.9% of patients had more than two cysts. Right lung involvement was present in 44.1% of the cases, left lung in 48.1% of cases; cysts were bilaterally located in 7.6% of the cases. The common lobes involved were right lower lobe (RLL) 31.0%, left lower lobe (LLL) 27.6%, left upper lobe (LUL) 24.1%, right upper lobe (RUL) 12.4%, and right middle lobe (RML) 4.8%.

Peripherally located cysts were more common than centrally located cysts (61.4% versus 27.6% respectively). We were not able to define the location for 16 cysts, which were large enough to reach both central and peripheral regions of lung. Calcification of cysts was only present in two cysts (1.4%). The lung was the solitary organ of the disease in 60.8% of the cases, whereas extrapulmonary involvement was detected in 39.2% of the cases. We detected one case with thymic involvement and three cases with multiple pleural cysts. The remaining cases had liver involvement. Airway compression was detected in 52.4% of the cysts.

Table 2. Radiological signs of the cyst rupture.

		n=69 cyst
Contained cyst rupture signs	Air crescent sign (%) Figure 1	15.9
	Inverse air crescent sign (%) Figure 2	2.9
	Air bubble sign (%)	5.8
Complete cyst rupture signs	Waterlily sign (%) Figure 3	24.6
	Cumbo sign (%) Figure 4	13.0
	Whirl sign (%)	15.9
	Dry cyst sign (%) Figure 5	14.5
	Mass within cavity sign (%)	13.0

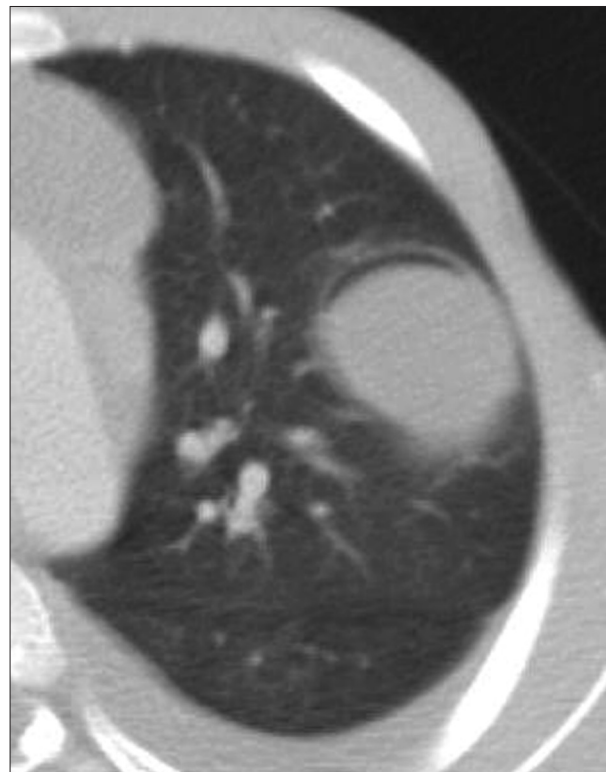


Figure 1. Air crescent sign. Air between the pericyst and the endocyst is compatible with contained cyst rupture in the left upper lobe seen on CT sections of a 9-year-old boy.

The prevalence of radiological signs of contained rupture and complete cyst rupture are given in Table 2. The most common sign was the waterlily sign with a prevalence of 24.6%. Contained rupture of a cyst was detected in 10.3% of cysts. The prevalence of complete rupture was 37.2%. Some cysts had more than one sign of rupture. In cases with presence of both signs of contained and complete rupture, the cyst was accepted as complete rupture. The overall number of ruptured cysts was 69 (47.5%), which was slightly fewer than intact cyst

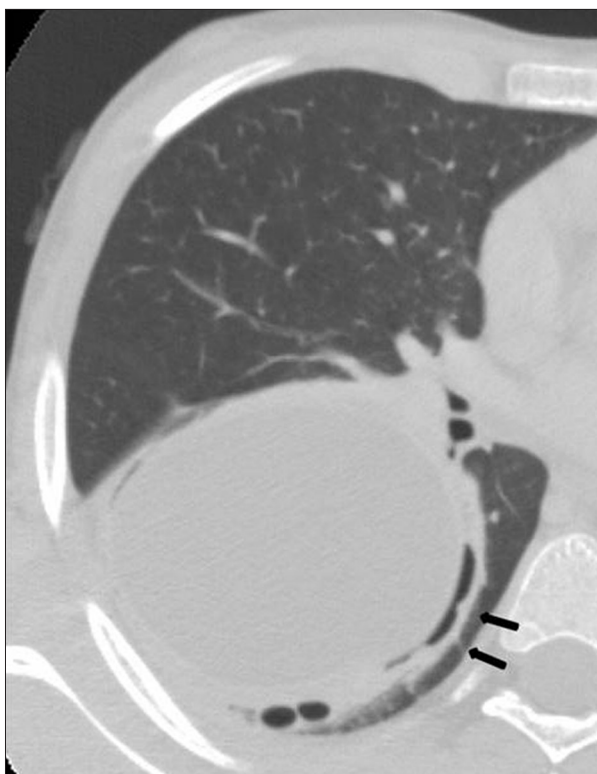


Figure 2. Inverse air crescent sign. Thin air density in posteromedial wall of cyst located in right lower lobe of a 15-year-old girl.

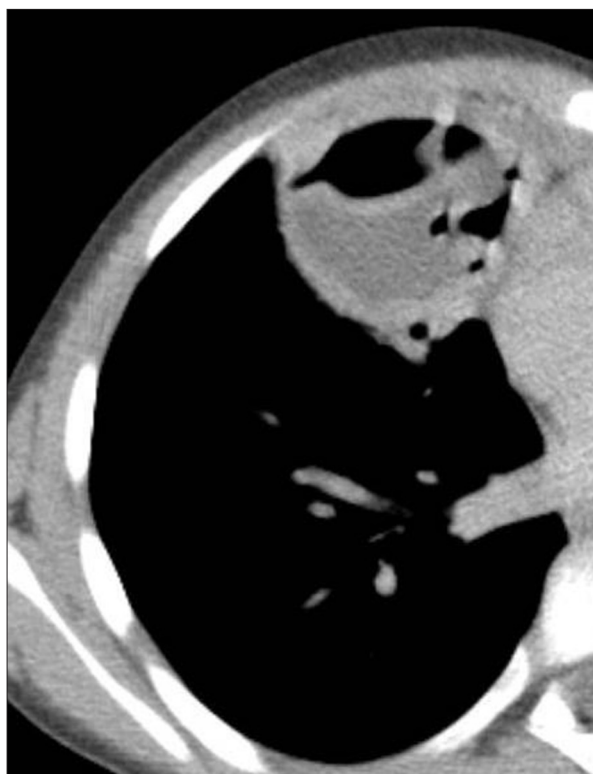


Figure 4. Cumbo sign. The air crescent causing detachment between the pericyst and endocyst and air fluid level within the endocyst in right medial lobe of 8-year-old girl.



Figure 3. Waterlily sign. Image of ruptured hydatid cyst in the right lung of an 8-year-old girl. Air-fluid level caused by bronchial rupture of cyst and generative membranes floating on the fluid can be seen.

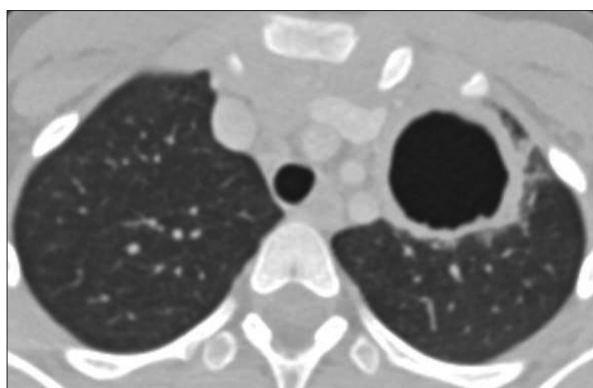


Figure 5. Dry cyst sign. The empty and air filled cyst is seen after expectoration of cyst content in ruptured hydatid cyst in left upper lobe of 9-year-old girl.

which was 76 (52.4%). Pleural rupture was detected in 3.4% of cases. The rupture rates were 42% in RLL, 57.1% in RML,

44.4% in RUL, 55% in LLL, and 40% in LUL. Rupture rate was 60% in peripheral cysts, 46% in central cysts, and 25% in undefined cysts ($p=0.054$). The side of the lung, the involved lobe, the number of cyst, the presence of extrapulmonary involvement, age, and sex were not different between the ruptured and non-ruptured cysts (Table 3). No cyst perforation was detected in cysts of 1 cm diameter. After exclusion of cyst of 1 cm diameter, the mean cyst diameter was 6.23 ± 2.83 cm in

Table 3. Comparison of ruptured and non-ruptured cysts.

	Ruptured cyst n=69	Intact cyst n=76	p
Age (years)	9.9±3.9	9.3±4.0	0.390
Sex (F/M) (n/n)	23/25	16/38	0.058
Mean cyst diameter (cm)	5.20±2.91	5.68±3.13	0.342
Side of lung (right/left/bilateral)	30/34/5	34/36/6	0.970
Involved lobe	RLL(n)	21	0.730
	RML(n)	4	
	RUL(n)	8	
	LLL(n)	22	
	LUL(n)	14	
Number of cyst	Single(n)	32	0.864
	2 cyst(n)	11	
	>2 cyst(n)	2	
Extrapulmonary involvement (%)	37.5	40.7	0.738
Central/peripheral (n)	24/41	16/48	0.143
Airway compression (%)	53.6	51.3	0.781

the unruptured group and 5.02±2.80 cm in the complete ruptured group ($p=0.020$). The mean cyst diameter was 6.38±3.11 cm in centrally located cysts, and 4.31±2.37 cm in peripherally located cysts ($p<0.0001$). Even after exclusion of perforated cysts, the mean cyst diameter was higher (7.12±2.85 cm) in centrally located cysts than in peripherally located cysts (4.27±2.50 cm) ($p<0.0001$).

The number of giant cyst with a diameter of 10 cm or longer was 26 (17.9%). Age, gender, side of lung, involved lobe, number of cyst, and extrapulmonary involvement were not different between the giant and non-giant cysts. Airway compression and percentage of centrally located cysts were significantly higher in the giant group than the non-giant group ($p<0.0001$, Table 4).

Discussion

Sex

Males constituted a high proportion of our study population (62% male versus 38% female). Similarly, most other studies indicated that males were more likely to be infected with pulmonary hydatid cyst disease in both children and adults [7,9,16]. The percentage of boys may be up 74% in infected children [17]. However, there was no sex differences reported in some adult and pediatric case series [17,18]. No studies with a predominance of infected girls have been reported. Boys may be more active outdoors in urban areas, which may

lead to a high probability of exposure to parasites and may be the cause of male predominance.

Site

We found that 70.6%, of the patients had a solitary cyst. Right lung involvement was present in 44.1%, left lung in 48.1%, and cysts were bilaterally located in 7.6% of the cases. The common lobes involved were right lower lobe 31.0%, left lower lobe 27.6%, and total lower lobes 57.6%. Our results were similar to literature reporting multiplicity of cysts in 30% of cases of pulmonary hydatid cysts, and similar to the predilection to location in the lower lobes of lung in 55–70% of cases [2,4,6–9,19,20]. However, cysts have been reported to be bilateral in 20% of cases, whereas in our study it was 7.6% of cases. Similar to our study results, the largest study that analyzing 1,155 pulmonary hydatid cysts detected bilateral pulmonary hydatid cysts in 7.3% of their cases [20].

Location

The central-peripherally distribution of pulmonary hydatid cysts has only been investigated in a few studies. The cysts in our study were more commonly peripherally located with a prevalence of 61.4%. Onal et al. recently reported peripherally location of cysts in 59.7% of 134 pediatric cases [9]. The mean cyst diameter was significantly higher in centrally located cysts than in peripherally located cysts (6.38±3.11 cm versus 4.31±2.37 cm $p<0.0001$) in our study. However, Kuzucu et al. [21] reported that centrally located cysts were

Table 4. The features of giant versus non-giant cysts.

	Giant cyst n=26 cyst n=24 patients	Non-giant cyst n=119 cyst n=78 patients	P
Age (years)	10.5±4.4	9.3±3.8	0.173
Sex (F/M) (n/n)	9/15	30/48	0.932
Mean cyst diameter (cm)	10.5±0.81	4.36±2.09	<0.0001
Side of lung (right/left/bilateral)	14/9/3	50/61/8	0.279
Involved lobe	RLL(n)	11	34
	RML(n)	3	4
	RUL(n)	3	15
	LLL(n)	5	35
	LUL(n)	4	31
Number of cyst	Single(n)	19	53
	2 cyst(n)	3	22
	>2 cyst(n)	2	3
Extrapulmonary involvement (%)	8/24	32/78	0.500
Central/peripheral (n)	12/5	28/84	<0.0001
Airway compression (%)	100	42	<0.0001
Rupture rate (%)	42.3	48.7	0.552

smaller than peripherally located cysts because of limited expansion of the cysts by the major bronchovascular structures in the central part of the lungs. Because perforation of a cyst may lead to shrinkage and decrease in diameter, we excluded perforated cysts, and reanalyzed our data. Centrally located cyst diameter continued to be higher than peripherally located cyst diameter in the re-analysis (7.12±2.85 cm versus 4.27±2.50 cm, $p<0.0001$). Our result, thus, conflicted with the previous a report by Kuzucu et al. [21]. Therefore, the issue needs to be clarified by new studies.

Giant versus non-giant

Giant hydatid cyst has been proposed as a distinct clinical entity. It is usually defined as a cyst with the largest diameter of more than 10 cm [5]. Giant cysts are reported to be more prevalent in children than adults [7,17]. The immature immune system and higher elasticity of the lung tissue may allow more rapid growth of the pulmonary hydatid cyst in children [5]. Prevalence of 15%–31% of giant cysts has been reported in pediatric case series [17,22].

In the comparison study between giant and non-giant cysts in 118 children, it was concluded that surgical need of parenchymal resection and postoperative complications were more common in patients with giant cysts [23]. In our study, the number of giant cysts with a diameter of 10 cm or larger was 26 (17.9%), which was similar to existing literature. In

a comparison of 26 giant cysts and 119 non-giant cysts, we found that centrally located cysts were significantly common in giant group ($p<0.0001$) and airway compression was present in all giant cysts, whereas the percentage was 42% for non-giant cysts ($p<0.0001$). However, no statistically significant difference was detected between giant and non-giant cysts.

Rupture

Rupture is the most frequent complication of pulmonary hydatid disease and may lead to significant clinical and radiological consequences. The cyst may rupture into the endobronchial system or into the pleural cavity. Ruptured cysts have been reported to represent greater postoperative morbidity and mortality than intact cysts [4,5,8,9] and in one study that included both adults and children, ruptured cysts occurred in more than half of 994 patients with lung cyst hydatid [20]. Another study found up to 60% complication rate for cysts in a population with adult and pediatric patients [24]. The prevalence of perforation of cysts was reported as 40%–60% in mixed populations [9,20,24]. Similarly, rupture rates of 33–45% have been reported in large pediatric studies [7,18,22]. The overall number of ruptured cysts was 69 (47.5%) in our study, which was slightly higher than previous reports.

Pleural rupture is reported in a wide range of pulmonary hydatid cyst disease, and ranges from 3.6–8.9% [9,10]. Similarly, pleural rupture was detected in 3.4% of our population.

However, a pleural complication rate up to 27.6% was reported by Aribas et al. [19].

A recent study found a significant difference between lobar location and rupture rate of cysts. The rupture rate was 70% in the right middle lobe and 66.7% in the lingual, and higher than in other lobes $p=0.018$ [9]. The presence of relatively smaller volumes in the right middle lobe and lingula, and compression by the chest wall and the heart are proposed as explanations of high rupture rates in these lobes. Although the highest rupture rate was detected, similar to a previous report, in right medial lobe (55%) in our study, the difference did not reach statistical significance.

There was no statistical difference between the peripheral/central locations of the cysts and the perforation rates [9]. The proportion of centrally located cyst was 34.7% in the ruptured group, which was higher than 21.0% in the non-ruptured group. There are controversial reports on the relationship of the diameter of the cyst and rupture of the cyst in the literature. Yalin et al. [13] investigated intracystic pressure (ICP) and the diameter of cysts in 50 abdominal hydatid cysts and concluded that as ICP increased, the diameter of the cyst also increased. However, that may not be the case for pulmonary hydatid cysts because of higher elasticity and low tissue resistance in lung compared to liver. Yuksel et al. reported that the mean diameter, volume, and ICP of 22 unperforated pulmonary hydatid cysts was 9.6 ± 4.2 cm, $728\pm 1,014$ cm³, and 36.6 ± 9.3 cm H₂O, respectively. They found no significant correlation between their short and long diameters, volumes, and ICP ($p>0.05$) [25]. Some authors reported [14] an enlargement in the size of the cyst as a negative risk factor for rupture. On the other hand, other authors [9,15] reported no relationship between the size of the cyst and the risk of rupture. The mean cyst diameter was 6.23 ± 2.83 cm in the unruptured group and was significantly higher than 5.02 ± 2.80 cm in complete ruptured group ($p=0.020$). The low diameter in the complete rupture group may be caused by shrinkage of the cyst after rupture. Therefore, shrinkage after rupture may blunt the relationship between rupture and radiologic size of cyst.

Signs of rupture

Communication of cysts with the bronchial tree shows a variety of more or less characteristic radiographic signs. Radiologic signs correspond to the different stages of the course of aging or damage of hydatid cysts [3,4]. Although typical imaging findings have been well described in the literature, radiologists should also be aware of atypical imaging findings that can occur secondary to rupture [6].

There are no papers comprehensively reporting all radiologic signs of contained and complete rupture separately.

Papers reporting prevalence of signs of rupture have used data from chest x-ray. Burgos et al. presented signs of cyst rupture of 70 patients where the lesion was an empty cavity in 28.6%, and the cyst was associated with pleural effusion in 11.4% [26]. Waterlily sign is seen in only 10–15% of lungs hydatid cysts [5,27]. In our study, the most common sign of rupture was the waterlily sign with a prevalence of 24.6% of ruptured cysts. We detected air crescent sign and whirl sign at a same prevalence of 15.9% in patients with ruptured cysts. We recommend searching all radiologic signs of contained and complete rupture in radiologic evaluations.

Extrapulmonary involvement

Concomitant involvement of pulmonary and hepatic hydatidosis is high, varying from 16–23.1% [5,7,18,26] in both children and adults. However, it may be higher in adults [5]. The number of co-involvements with the liver may not reflect the exact number because not all patients underwent abdominal imaging. In some patients we were only able to investigate liver sections seen on thoracic CT. This may be a limitation of our study.

Calcification

Calcification in pulmonary cysts is rare (0.7% of cases) [4]. However, Burgos et al. reported cyst calcification in 4.3% (4/70 ruptured cysts) of pulmonary hydatid cysts [26]. In our study, calcification of cysts was only present in 2 cysts (1.4%). Unlike extrapulmonary cysts, pulmonary hydatid cysts do not undergo calcification, and daughter cyst formation is also rare. Low carbon dioxide content within the lung parenchyma reduces the serum calcium level despite the presence of sufficient phosphorus released by tissue necrosis for calcium precipitation [5].

Conclusions

Pulmonary hydatid cysts commonly presents as solitary cysts with a predilection of lower lobes and peripheral regions of the lung. One third of patients may have concomitant extrapulmonary involvement. Peripherally located cysts were more common, but smaller than centrally located cysts. Inconclusive results about cyst diameter and rupture may be partly caused by shrinkage of cysts after rupture. Radiologists should also be aware of atypical imaging findings that may be caused by rupture, and radiologic signs of contained and complete rupture should be explored in radiologic evaluations. The relationship of cyst rupture and size and location warrants further studies.

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