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Imaging In 100 Patients of Thoracic Hydatid Disease Including Unusual Imaging Appearances

Background/Objective: To evaluate the chest radiography and CT scan characteristics of pulmonary hydatid disease (PHD).

Patients and Methods: One hundred patients (59 males and 41 females, age ranged from 9 to 80 years) with surgically proven pulmonary hydatid cysts were studied. We reviewed clinical and imaging findings including PA and LAT chest roentgenograms and conventional CT of the chest. Only 82 patients had CT scan in their files, but all had CXR. The radiological features (localization, diameter, architecture, density and other radiological signs and appearances) were determined.

Results: On CXR, 124 cysts were determined. In evaluation of 82 available CT scans, a total of 112 cysts were detected. No cysts was detected on 5 CT scans. No discrete cyst was detected on 10 CXRs: 4 patients, only consolidation; and 6 patients, only hydropneumothorax.. The most frequent site of involvement was RLL (29.6%). Fifteen hydatid cysts appeared as solid masses on CT. Fifty-seven cysts were ruptured cysts and 25 patients with ruptured cysts had hemoptysis (43.9%). Thirty-eight percent of cysts had thin walls and 62% had thick walls. Sixty-four cysts were round in shape (55.7%). Single cysts were seen in 63 patients while multiple cysts were seen in 37. Median CT density of the cysts was 24 Hounsfeild Units (HU) (-18 to 84). There were 16 giant cysts (diameter $\geq$ 10 cm) on CT. Mean maximum and minimum dimensions of cysts were 5 cm and 4 cm on CT and 6.8 cm and 5.7 cm on CXR, respectively. On CT and CXR, "water lily sign" was seen in 18 and 22 patients, "air-fluid level" in 12 and 17 patients, and "crescent sign" in 11 and 5 of patients, respectively. Inverse crescent sign and calcification were not observed on CXRs, but each was reported on 4 CT scans. On CT, 90% of cysts were smooth, 74 cysts were uniloculated and 9 were multiloculated. Nineteen percent of cysts were infected. Other imaging findings included mediastinal shift, atelectasis, infiltration, pericystic lung reaction, chest wall involvement, and rib destruction.

Conclusion: CXR is helpful with diagnosis of intact cysts but fails to define entire morphology of complicated cysts. CT imaging recognizes certain details not visible on radiography. In endemic regions like Iran, atypical imaging presentations of complicated pulmonary hydatid disease, such as solid masses, should be considered in differential diagnosis of pulmonary lesions

Keywords: pulmonary hydatid disease, imaging

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Introduction

Hydatid disease is a parasitosis and is endemic in many sheep-rearing regions in the world, especially in the Middle East and Mediterranean countries. [1–3] The reported annual morbidity from the disease in humans is 1.04–2.4 per 1,000,000, while the actual rates are supposed to be more than twice that due to underreporting .[4] Lungs are the most common sites of infection in children, while liver is the most common site affected in adults. [2]

Almost all symptoms of hydatid disease are due to the pressure effect on surrounding structures, resulting from distension, obstruction, erosion, or infection.[5] Diagnosis is based upon immunodiagnostic tests and liver imaging findings. Conventional radiography is routinely employed for diagnosis of the disease in developing countries. CT, however, has been available only recently in many of

such places; as such, the CT findings of hydatid cysts of the lung are only sparsely described. [6, 7]

Patients and Methods

Patients

In this study, we searched for patients with surgically proven diagnosis of pulmonary hydatid disease (PHD) in whom pathological examination had confirmed hydatid disease. Records of 100 pationts (59 males and 41 females, aged 9 to 80 years) with surgically proven pulmonary hydatid cysts were reviewed retrospectively. Patients had been hospitalized in the Thoracic Surgery Ward of Masih Daneshvari Hospital (NRITLD) between 2002 and 2004. Eighty-one patients (81%) had not used oral drugs for hydatid disease before surgery. We reviewed clinical and imaging findings of including posteroanterior and lateral chest roentgenograms and conventional CT of the chest.

The most common symptoms of patients were cough, sputum, dyspnea, chest pain and hemoptysis (Table 1). Laboratory tests, especially complete blood count (CBC) and differentiatl, were evaluated. Eosinophils normally constitute 1 to 3 percent of peripheral blood leukocytes.[8] If eosinophils comprised more than 2.5 percent of WBCs, test of eosinophilia was considered positive.

Imaging technique

All patients had chest radiography before surgery, obtained at 80 KVp using CGR 1000 mA.

Only 82 patients already had CT scan in their files. Computed tomography had been performed with the same CT scanner using the sequential technique (collimation 10mm, spacing 10mm) with Siemens Somatom plus (Elinger, Germany). CT window widths ranged form 1300-1500 Hounsfield Units (HU) and window levels ranged form 500 to 600 HU in the parenchymal view. Also, window widths and levels ranged in mediastinal view between 350-400 HU (ww) and 40-50 HD (wl), respectively. Two board-certified radiologists reviewed chest radiographies and CT scans. Radiological features (localization, diameter, architecture, density and other radiological signs and appearances) were determined. Finally, the additional information obtained from CTs and CXRs were recorded.

Statistical analysis was performed by SPSS 11.5 for Windows.

Radiological signs and special features

-*Crescent sign (meniscus sign)*: a crescent-shaped air shadow toward the top of cyst (produced by introduction of air between the pericyst and endocyst).[9]

-*Inverse crescent sign*: a crescent-like rim of air observed at the lower end of cyst with appearance that was morphologically opposite the classic "crescent" sign.[9]

-*Water lily sign*: endocyst membrane floating on top of the remaining fluid, subsequent to collapse of endocyst and partial evacuation of fluid.

-*Infected cysts:* poor delineation, air-fluid or fluid-fluid levels,[10] pericystic lung parenchymal reaction and wall thickening (wall thickness ≥ 3 mm).

Operation

Posterolateral thoracotomy and cyst resection was performed for each patient. Forty-one patients had 2-year follow-up with CT scan, radiography and sonography, of which 32 patients did not show recurrent hydatid disease.

Results

On CXRs, 124 cysts were noticed. No discrete cyst was detected on 10 CXRs, and Radiographic findings in this group were as follows: 4 CXRs, only consolidation; 6 CXRs, only hydropneumothorax. Of 82 available CT, these was no cayst on 5 CT scans: 112 cysts were detected on 77 of the CT scans. Fifteen hydatid cysts appeared as solid masses. Seventy-four cysts were uniloculated and 9 were multiloculated. Fifty-nine cysts were ruptured (air having dissected into the cyst causing separation of the membranes), while 53 were unruptured.

Other organ involvement

Abdominal ultrasound had been performed in 73 patients (73%). Of 73 abdominal ultrasounds, 49 were normal (68%) but 20 showed liver involvement with hydatid cyst (27%), one showed spleen involvement (1%) and 3 (4%) showed both liver and spleen involvement.

Relation with symptoms

As mentioned before, the most frequent presenting symptoms of patients were cough (67%), sputum (53%), hemoptysis (38%), chest pain (38%) and dyspnea (36%) (Table 1). Totally, 7 patients were asymptomatic. Fifty-seven patients had at least one ruptured cyst and 43 had no ruptured cysts. Hemoptysis was present in 25 cases with ruptured cyst (43.9%), although only 13 cases with intact cysts had hemoptysis (30.2%). Only 3 patients with ruptured cyst were asymptomatic.

Laboratory tests before surgery in 70 patients (70%) did not show eosinophilia, although 30 patients (30%) were positive for it. On CT scans of 21 patients, infected cysts were detected (27.2%). It is notable that fever was the presenting sympton in 22% of patients.

Table 1. Presenting symptoms.

Symptoms	Patients No.	%
Cough	67	67
Sputum	53	53
Hemoptysis	38	38
Chest pain	38	38
Dyspnea	36	36
Fever	22	22
Weight loss	14	14
Abdominal pain	8	8
Weakness	2	2
Night sweet	1	1
Itching	1	1

Site of involvement

All lung lobes were involved. The most frequent site of involvement was the right lower lobe (RLL), and the least frequent one was the lingula (Table 2).

Table 2. Site of cysts involvement (RLL: right lower lobe, LLL: left lower lobe, LUL: left upper lobe, RUL: right upper lobe, RML: right middle lobe, No.; number of cysts).

Site	No.	%
RLL	37	**29**
LLL	35	**28**
LUL	18	**15**
RUL	16	**13**
RML	12	**10**
Lingula	**6**	**5**

Number of cysts

Of 90 patients with cysts on chest x-ray, 66 patients (73.3%) had a single cyst, and others had multiple cysts (2-7 cysts). On 82 available CT scans, 112 cysts were detected, of which 57 were single and others were multiple (Table 4).

Table 3. Number of lung cysts detected on chest x-rays and CT scan. Cyst number with frequency and percentage of patients are shown (No.: number of cysts, Fre: frequency of patients, Per: percent of patients with recorded cyst on CXR or CT).

	CXR		CT	
No of cysts	Fre	Per	Fre	Per
1	66	73.3	57	74
2	16	17.7	14	18.1
3	6	6.6	3	3.8
4	2	2.2	0	0
5	-	-	1	1.2
6	-	-	1	1.2
7	-	-	1	1.2

Other CT findings

Pleural effusion, pneumothorax, hydropneumothorax, collapse, and laminated membrane were other CT findings (Table 4).

Table 4. CT findings in patients without any discrete cyst on CT

Patient code	CT findings
1	Pleural effusion with floating laminated membrane
2	Pleural effusion, pneumothorax, collapse of lingula
3	Pleural effusion with laminated membrane (water lily sign)
4	Pleural effusion, right hydropneumothorax
5	Fistula from liver, right lower lobe consolidation

Wall thickness

Wall thickness of 50 cysts on CT scans and 40 cysts on CXR could be measured (Table 5).

Table 5. Wall thickness on CT and CXR (No.: number of cysts).

	CT		CXR	
	No.	%	No.	%
Thick_ walled	31	62	23	57.5
Thin _walled	19	38	17	42.5

Cysts shape on CT

The shapes of the cysts were round (most common), oval, irregular, and pear-shaped (the least frequent one) (Table 6).

Table 6. Shapes of cysts on CT.

Shape	Count	%
Round	64	57
Oval	28	25
Irregular	19	17
Pear-shaped	1	1

Density

Median CT density of cysts was 24 Hounsfeild Units (HU) (-18 to 84).

Size

Mean maximum and minimum dimension of cysts were 5 cm (range: 1-15) and 4 cm (range: 1-12) on CT and 6.8 cm (range: 1-19) and 5.7 cm (range: 1-13) on CXR, respectively. There were 16 giant cysts (≥ 10 cm of cyst diameter on CT).

Radiological signs and findings

Of respectively 112 and 124 detected cysts on CT and CXR, water lily sign was observed in 18 and 22 cysts, air-fluid level in 12 and 17 cysts, and crescent sign in 11 and 5 cysts. Inverse crescent sign (a crescent-shaped rim of air at the lower end of the cyst) [9] and calcification were not observed on CXRs, but each one was recorded in 4 cysts on CT scans (Table 7).

Other imaging findings were pleural effusion, hydropneumothorax, mediastinal shift, atelectasis, infiltration, pericystic lung reaction, chest wall involvement and rib destruction.

Of 100 radiography films and 82 CT scans, pleural effusion was seen on 15 CXRs and 21 CTs. Fifteen CXRs showed hydropneumothorax (Figures 2 and 3), and 16 showed infiltration. Seven CXRs showed mediastinal shift, although this finding was recorded only on 4 CTs. Atelectasis was the finding on 2 and 1 CXRs and CT, respectively. Overall, 30 patients had pleural involvement (30%) including pleural effusion, pleural thickening, pneumothorax or hydropneumothorax.

Of 112 detected cysts on CT, pericystic reaction in

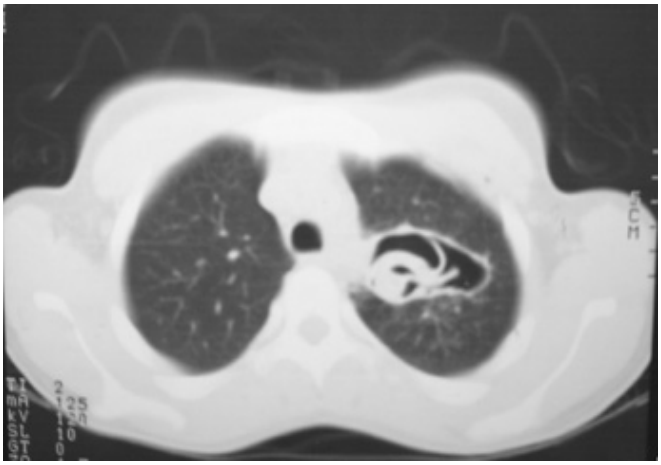

Fig 1. Cavitary lesion with visible laminated membrane

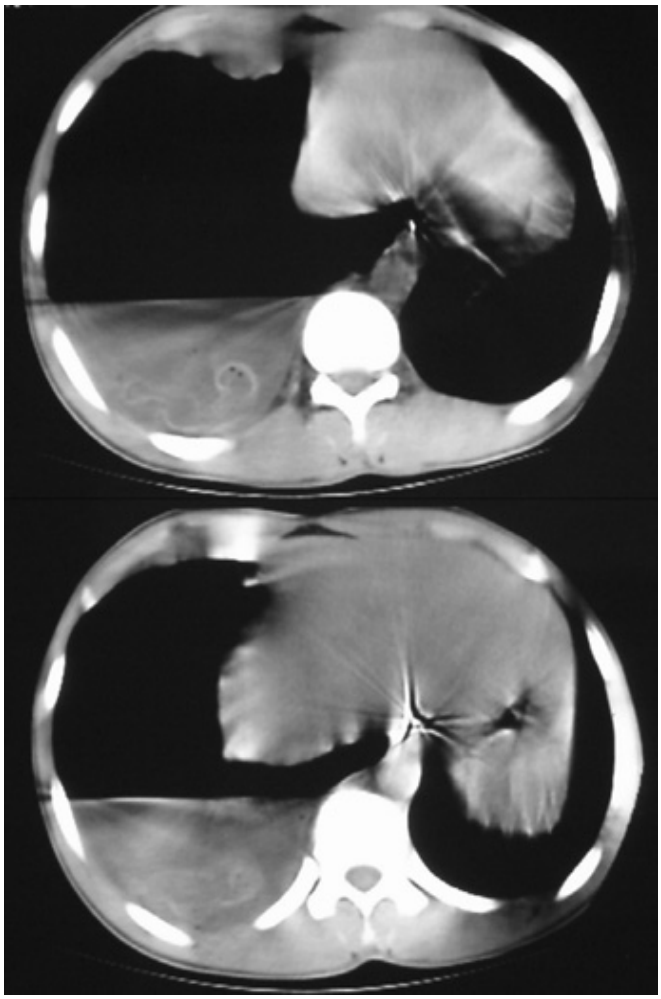

Fig 2. Large right sided hydropneumothorax, with visible laminated membrane in pleural fluid

Table 7. Radiological findings detected on CT scan and CXR (No.: number of cysts,)

Rad Fin	Water lily sign		Air-fluid level		Crescent sign		Inverse crescent sign		Calcification	
	CXR	CT	CXR	CT	CXR	CT	CXR	CT	CXR	CT
No.	22	18	17	12	5	11	0	4	0	4
%	17.7%	16%	13.7%	10.7%	4.4%	8.8%	0.00%	3.5%	0.00%	3.5%

the lung tissue was repored in 31 cysts. Only one patient had chest wall invasion and rib destruction detected on his CT and CXR (Figure 4). One patient had hepatobronchial fistula that referred with cough and abdominal pain and the finding on his chest x-ray was a mass consolidation in the right lower lobe.

Discussion

Hydatid disease has been known since the time of Hippocrates. The clinical features of the disease have been described since then. Hydatid disease is endemic in some parts of the world including Iran. It is still an important public health problem, especially in rural areas of western regions of Iran (Figure 5). Hydatid disease is seen in any age and sex group, although it is more common in 20-40 years of age. [11-13] The results of our study were in harmony with those in the literature: the male/female ratio was 1.43 and the mean age of the patients was 32.11 years; the median age was 30 years. Of 100 patients in our study, 45 were in the range of 20 to 40 years.

In humans, hydatid disease involves the liver in approximately 75% of cases, lungs in 15%, and other anatomic location in 10%. [14-17] The lungs are the second most frequent site of hematogenous spread in adults and probably the most common site in children (15-25% of cases). [16, 18] Jerray et al. in a study of 386 cases and Dogan et al in a study of 1055 patients concluded that sudden coughing attacks, hemoptysis, and chest pain were the most common clinical symptoms .[11, 19] As noted before, the most frequent symptoms in our study were cough (67%), sputum (53%), hemoptysis (38%), chest pain (38%) and dyspnea (36%). The most frequent symptoms in our study were compatible with these two large studies. Many hydatid cysts, however, are asymptomatic and are found incidentally on chest radiographs. [20]

According to previous articles, cysts are multiple in 30% of cases, bilateral in 20%, and located in lower lobes in 60%. [10,16,1,19] In our study, the most frequent sites of involvement were lower lobes (right: 37 cysts (29%), left: 35 cysts (28%)). In a review article, Pedrosa et al. showed the predominance of lower lobes involvement. [17]

The number of cysts can be as high as 60 and concurrent involvement of the liver and lungs is seen in approximately 6% of all patients with cysts in thoracic and abdominal organs. [10, 21] In our study, liver involvement was present in 27% of patients and the highest number of lung cysts was 7.

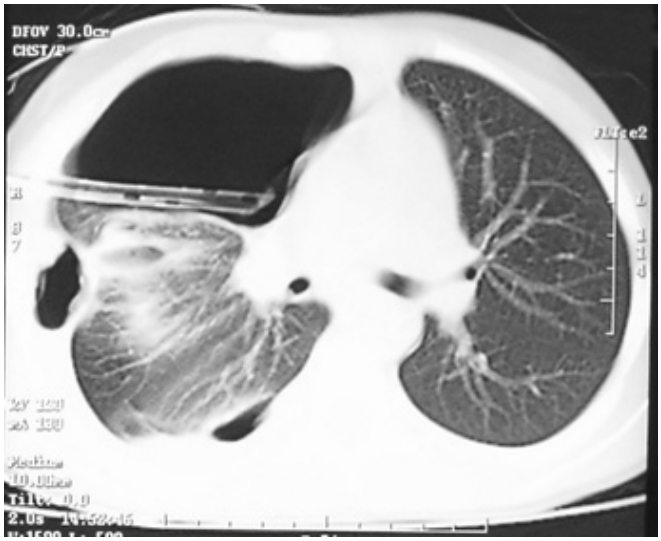

Fig 3. Right sided hydropneumothorax with chest tube in place. No discrete cyst or laminated membrane was visualized.

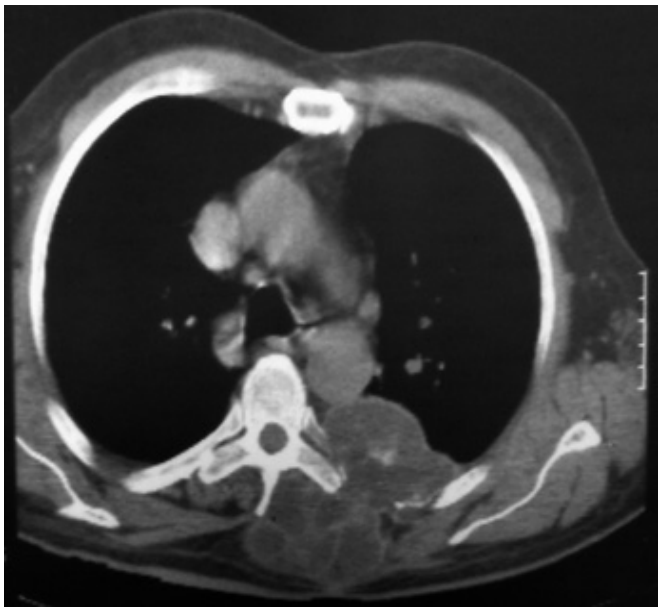

Fig 4. Multiloculated cystic mass in Lt paravertebral region with chest wall extension and rib erosion.

Radiologically, pulmonary hydatid cysts are classified into three types on the basis of their appearance: simple cyst (with no internal architecture), calcified cyst, and complicated cyst.

Conventional chest radiography has been the mainstay of diagnostic armamentarium for patients with hydatid disease of the lungs, often coupled with

Casoni's skin testing and serologic testing for antibodies. However, the two, even in combination, may not yield a definitive diagnosis as a result of many conditions, such as carcinoma, benign tumor, inflammatory mass, metastasis, and solid or fluid-filled cyst mimicking the radiologic features of hydatid cysts of the lung. [9]

Even though chest roentgenogram has been considered in previous articles as a fairly diagnostic method, hydatid lung disease may present in a variety of radiological forms, and it is particularly impossible to define the entire morphology of complicated cysts. [11, 22-25] CT scanning has come to the rescue of clinicians in elucidating the cystic nature of the lung mass and accurate localization for planning surgical treatment .[9] The present study supports this view even for developing countries. In this study, we have reviewed imaging findings in cases of pulmonary hydatid disease.

Hydatid cyst complications include rupture and superinfection. Rupture occurs in 50-90% of cases and includes rupture of the cyst into parenchyma and pleural cavity. Cyst rupture is mainly due to degeneration of parasitic membranes as a result of age, chemical reaction, or host's defense mechanisms. [10] Cyst growth produces erosions in the bronchioles that are included in the pericyst, and as a result, air is introduced between the pericyst and the laminated membrane.[16] This air collection appears as a thin, radiolucent crescent in the upper part of the cyst and is known as the crescent sign or the meniscus sign.[16,19, 26] Some authors consider this to be a sign of impending rupture and an indication for emergency thoracotomy.[16] As air continues to enter this space, the two layers separate completely and the cyst shrinks and ruptures, allowing the passage of air into the endocyst.[16,26] When it has completely collapsed, the crumpled endocyst floats freely in the cyst fluid (the water lily sign) (Figure 1). [19,25,27] If the fluid is entirely evacuated by expectoration, the remaining solid components will fall to the most dependent part of the cavity ("mass within a cavity"). In our study, 18 cysts appeared as cavitary lesions.

Cysts may rupture directly into pleural and peritoneal cavities. According to a review article by Plat et al. up to 25% of ruptured cysts may become infected. [10] In our study, ruptured cysts detected on CT were 52%. The mural characteristics of pulmonary hydatid cyst have been reported earlier by Koul et al. for the first time in 2000.[9] The reason for the thickness could

Fig 5. Geographic distribution of hydatid disease. The map shows areas in which hydatid disease is endemic due to the transmission of E. granulosus through the dog-sheep cycle (solid red areas). Red stripes indicate areas where transmission occurs by means of alternative life cycles in which carnivores such as wolves and foxes serve as definitive hosts and goats, camels, and horses serve as intermediate hosts. Transmission by means of alternative life cycles is common in North Africa, the Middle and Far East, the United States, Canada, and Iceland.
[adopted from "Principles and practice of infectious diseases" 4th ed]

be attributed to the infection of the cysts, which was observed in all the 31 cysts with thick walls on CT. Our study showed CT density of cysts ranging from -18 to 84 Hounsfeild Unit (median: 24 HU) although it ranged from -42 to 160 Hounsfeild Units in Koul PA et al's study (median: 15.5 HU). [9] Also in that study, the size of cysts varied from 1.5 to 13 cm. In our study, the range of cyst size was 1-15 cm on CT.

Calcification in pulmonary HC is very rare (0.7%), yet these were four calcified cysts (3.5%) on CT scans in our study. Calcification manifests as round, hyperattenuating areas on CT. [10]

Although it is rarely seen, a more serious type of rupture is the cyst perforation into pleural or pericardial cavities. [12, 24] The occurrence of these complications is the result of the rupture of cysts located in lungs or in the dome of the liver. [11, 24] Our results regarding pleural involvement were similar to Aribas's study, while pleural complications are reported to range between 0.5% and 18.2% in the literature.[11] It was interesting that in patients with pulmonary hydatid disease, there was no discrete cyst on 5 CTs and 10 CXRs. And also, chest wall involvement (muscles and ribs) was seen in one patient (Figure 4). One patient (1%) showed hepatobronchial fistula in our study, although Aribas et al recorded this finding in 7% of patients.[11]

Conclusion

Chest radiography is helpful for diagnosing of intact cysts but it is unable to define the details of morphology of the complicated cysts. CT imaging, if available, can recognize certain details of the lesions and discover others not visible on radiography. In endemic regions like Iran, atypical imaging presentation of complicated pulmonary hydatid disease, such as solid masses, should be considered in the differential diagnosis of pulmonary lesions.

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