СЛУЧАЙ ИЗ ПРАКТИКИ

PULMONARY ECHINOCOCCOSIS WITH EXTENSION TO SPLEEN AND LIVER IN A PATIENT WITH HAIRY CELL LEUKAEMIA AT THIRD RELAPSE

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Human cystic echinococcosis is a widely endemic helminthic disease transmitted by dogs in livestock raising areas. It is a widely endemic helminthic disease worldwide in which humans are infected as 'aberrant' hosts by Echinococcus granulosus and develop cysts in numerous different organs. Clinicians, radiologists and pathologists should be aware of this entity and its pulmonary manifestations. Depending on their size and anatomic location, the cysts can eventually exert pressure on adjacent structures and be associated with unspecific symptoms like chronic cough, dyspnoea, pleuritic chest pain and haemoptysis. Differential diagnoses in the lungs include primary lung carcinoma and metastatic disease; related complications include pleuritis, lung abscess and pneumothorax.

This report presents the imaging findings on chest X-rays and tomography of cystic echinococcosis in the liver, lungs and spleen in a patient with hairy cell leukaemia at third relapse. Images depict the initial diagnosis and six-month follow-up. We supplement the case with a brief literature review.

Keywords: lung echinococcosis, hydatid disease, differential diagnosis, frozen section.

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ЭХИНОКОККОЗ ЛЕГКИХ С РАСПРОСТРАНЕНИЕМ НА СЕЛЕЗЕНКУ И ПЕЧЕНЬ У ПАЦИЕНТА С ВОЛОСАТОКЛЕТОЧНЫМ ЛЕЙКОЗОМ ПРИ ТРЕТЬЕМ РЕЦИДИВЕ

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Кистозный эхинококкоз человека – широко распространенное эндемичное гельминтозное заболевание, передаваемое собаками в районах животноводства. Это широко распространенное гельминтозное заболевание во всем мире, зооноз, при котором люди являясь случайными промежуточными хозяевами, заражаются Echinococcus granulosus и у них развиваются кисты в различных органах. Клиницисты, рентгенологи и патологоанатомы должны знать об этом заболевании и его легочных проявлениях. В зависимости от размера и анатомического расположения, кисты могут оказывать давление на соседние структуры и быть связаны с такими неспецифическими симптомами, как хронический кашель, одышка, плевральная боль в груди и кровохарканье. Дифференциальный диагноз включает первичную карциному легких и метастатическое поражение легких; сопутствующие осложнения включают плеврит, абсцесс легкого и пневмоторакс.

В данном наблюдении представлены результаты рентгеновского исследования грудной клетки и компьютерной томографии кистозного эхинококкоза в печени, легких и селезенки у пациента с волосатоклеточным лейкозом при третьем рецидиве. Изображения демонстрируют первичное исследование и период после шести месяцев наблюдения. Случай дополнен кратким обзором литературы.

Ключевые слова: эхинококкоз легких, кистозный эхинококкоз, дифференциальная диагностика, криостатный срез.

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Introduction.

Human cystic echinococcosis is a widely endemic helminthic disease transmitted by dogs in livestock raising areas [1]. Humans are infected as 'aberrant' hosts by Echinococcus granulosus and develop cysts in numerous organs. In humans, cysts may develop in multiple anatomic sites. This form of echinococcosis is termed primary cystic echinococcosis (CE). Secondary CE, predominantly in the abdominal and pleural cavity, results from spontaneous or trauma-induced cyst rupture and the release of protoscoleces and small cysts, which can grow to larger cysts [2, 3]. Approximately 40-80% of patients with primary CE have single-organ involvement and harbour a solitary cyst. Hydatid fluid is antigenic; thus, immune-mediated reactions such as urticaria, asthma, membranous nephropathy and anaphylaxis can occur [4, 5]. Because oncospheres enter the circulation via the gastrointestinal tract, the most common site of this disease in humans is the liver (50-70%), followed by the lungs (20-30%), and less frequently other organs including kidneys, spleen, muscles, skin, abdominal and pelvic cavities [3, 6]. Due to the

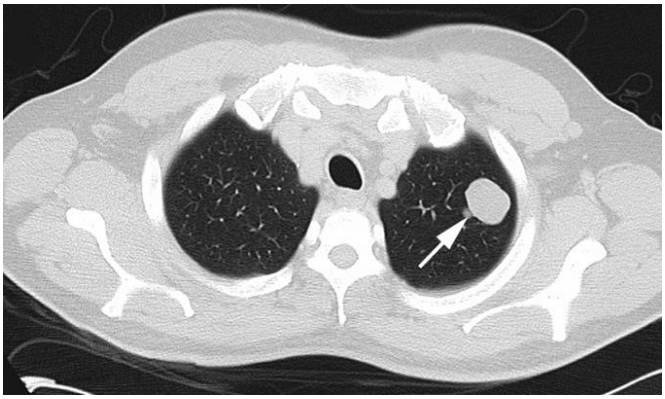
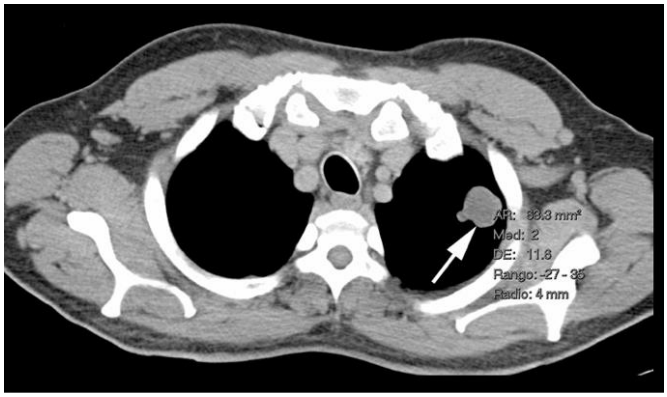


Fig. 1 a (Рис. 1 а)



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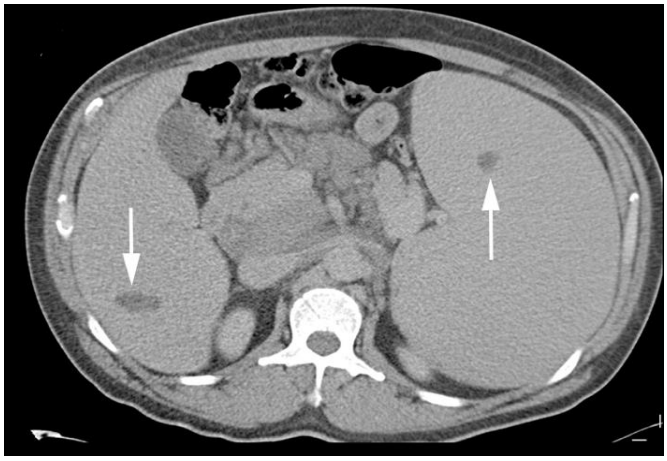

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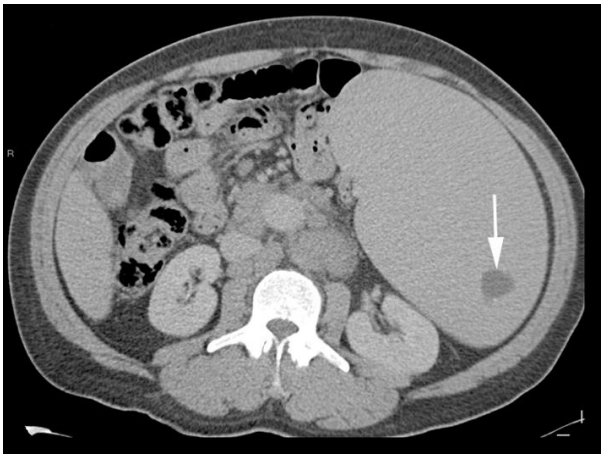


Fig. 1 d (Рис. 1 г)

Fig. 1. CT, axial view. A, B – chest tomography showed a pulmonary nodule with dimensions of 2x3 cm in the left upper lobe (white arrows). C, D – the abdominal tomography showed hepatomegaly and splenomegaly with cystic-like lesions in the right liver lobe and spleen (white arrows). rior quadrants.

Рис. 1. КТ, аксиальная плоскость. A, B – КТ органов грудной клетки, определяется легочный узел размером 2x3 см в левой верхней доле (белые стрелки). C, D – КТ органов брюшной полости; гепатомегалия и спленомегалия с наличием кистозных образований в правой доле печени и селезенке (белые стрелки).

slow growth of cyst development, CE can go undetected for years. Cysts can eventually exert pressure on adjacent structures depending on their size and anatomic location. They can be associated with unspecific symptoms, and when in the lungs, they might provoke a chronic cough, dyspnoea, pleuritic chest pain and haemoptysis [1].

This report presents a patient with cystic echinococcosis with evidence of cysts in the liver, lungs and spleen. We comment on the imaging findings on chest X-rays and tomography and the follow-up with a brief literature review.

Case presentation.

On day one of this report, a 52-year-old male consulted the outpatient clinic of our hospital complaining about fever, inspiratory stridor, and abdominal pain during the preceding two weeks. The patient's medical history refers to a 17-year-old diagnosis of hairy cell leukaemia, currently in its third remission. On physical examination, the patient had a fever over 38oC, splenomegaly and hepatomegaly, and pulmonary rales in the left lung; laboratory studies showed pancytopenia (600 leukocytes) and laryngeal infection by pseudomonas. Due to these findings, the patient underwent chest and abdominal tomography.

Chest tomography revealed a smoothly delineated cystic tumour with dimensions of 2x3 cm in the left upper lobe (Fig. 1 A-B). The tomography showed hepatomegaly and splenomegaly in the abdomen with five cystic lesions in the right liver lobe and 4 in the upper and lower poles of the spleen (Fig. 1 C-D, 2 A-D). The remainder parenchyma in the lungs, liver and spleen appeared inconspicuous.

Radiologically, either a tuberculous cavern or echinococcosis was suspected. Bronchoscopy revealed no pathological changes. PCR could not

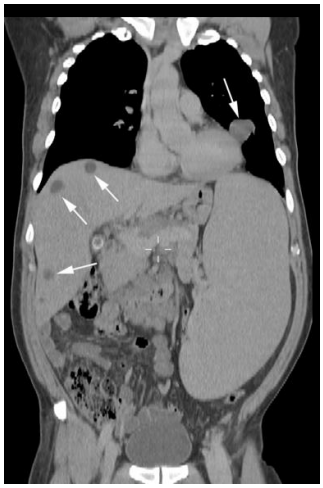


Fig. 2 a (Рис. 2 а)

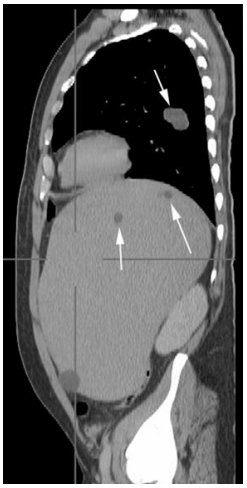


Fig. 2 b (Рис. 2 б)

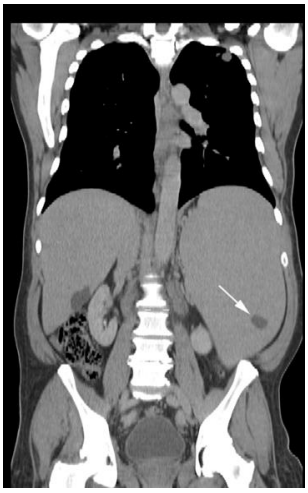


Fig. 2 c (Рис. 2 в)

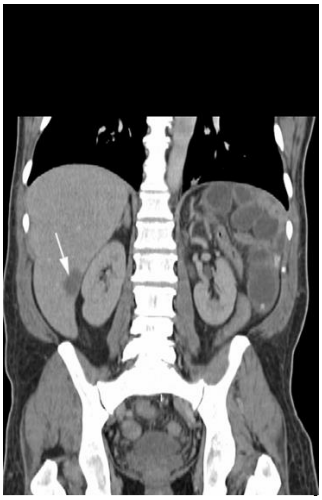


Fig. 2 d (Рис. 2 г)

Fig. 2. CT, abdomen, multiplanar reconstructions. A-D – multiplanar reconstruction confirmed the hepatomegaly and splenomegaly with evidence of 5 cystic lesions in the right liver lobe and at least four cystic lesions in the upper and lower poles of the spleen (white arrows).

Рис. 2. КТ органов брюшной полости, мультипланарная реконструкция. A-D –гепатомегалия и спленомегалия с признаками 5 кистозных поражений в правой доле печени и не менее четырех кистозных поражений в верхнем и нижнем полюсах селезенки (белые стрелки).

verify a mycobacterial infection in the serum or bronchoalveolar washing.

Due to the clinical and imaging findings, the patient was hospitalised, antibiotics were started with imipenem, vancomycin, and caspofungin, and scheduled splenectomy was recorded. He continued his management and surveillance by the haematology, oncology, and surgery services. The exudate culture results confirmed the diagnosis of tracheitis due to Pseudomonas aeruginosa.

On days 33 (Fig. 3A) and 35 (Fig. 3B), the patient had a chest X-ray that showed evidence of pneumonic foci, with a reticular pattern in the lower right lobe, with proof of already known cystic lesions in the lower left lobe (Fig. 3). The patient received antibiotic treatment with ceftriaxone and clarithromycin and was later switched to piperacillin and tazobactam when the presence of tracheitis was due to Pseudomonas aeruginosa was documented.

A chest tomography on the same day, 35, showed areas of pneumonic consolidation in the left upper lobe (Fig. 4A). Because serological testing for Echinococcus species was negative, a CT-guided biopsy of one of the cystic lesions was performed on day 39 and submitted to the Department of Pathology for diagnostic work-up; it confirmed the diagnosis of pulmonary hydatidosis (Fig. 4B).

The patient had splenectomy and biopsy of liver and lung lesions on day 50 of admission. On day 60, a histopathological diagnosis of hydatidosis was made for liver and spleen lesions, in addition to splenic infiltration due to leukaemia; This confirms the diagnosis of disseminated hydatidosis to the liver, lung, and spleen. Because the size of the cystic lesions was not greater than 5 cm, no surgical intervention was planned, and only medical treatment was continued. So he receives treatment in the infectology area with 400 mg albendazole every 12 hours for three months.

On day 174, 4.5 months later, a control chest CT scan showed an area of atelectasis in segment IV of the left lower lobe (Fig. 4C) and the presence of air in the already known cystic lesion. On day 225, 7.5 months after admission, a control tomography showed the resolution of the cystic lesions of the left lung. However, sequelae of areas of pleural thickening were observed in the left upper lobe (Fig. 4D and 4E); Also in the basal region of the lower left lobe, areas of pleural thickening were observed, which conditioned retraction of the cardiac silhouette, with the presence of a residual area of pulmonary consolidation and an air bronchogram (Fig. 4F).

The patient resections of adhesions and cystic lesions in pulmonary segments 3, 6, 8 and 9; of pericardial lingula and anterior chest wall on day 182 after his admission. After surgery, the patient did not have respiratory distress; it was 95% saturated. On day 188, the diagnosis of pulmonary tuberculosis was confirmed. Eventually, he was discharged and continued antibiotic

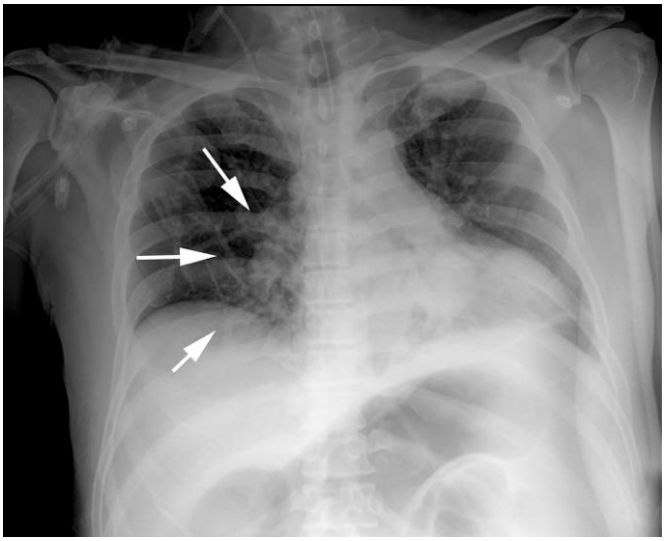


Fig. 3 a (Рис. 3 а)

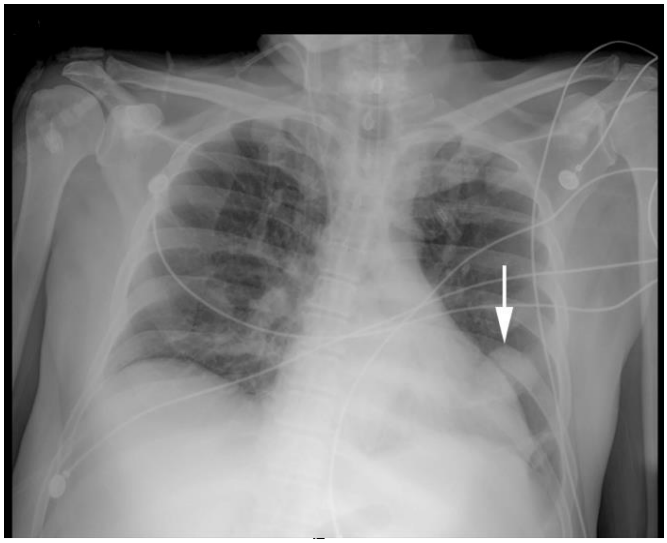


Fig. 3 b (Рис. 3 б)

Fig. 3. Chest X-rays, PA view. X-rays were taken on days 33 (A) and 35 (B) of hospital stay, showing the appearance of pneumonic foci, with a reticular pattern in the lower right lobe, with evidence of already known cystic lesions in the lobe lower left (white arrows).

Рис. 3. Рентгенограммы органов грудной клетки, прямая проекция; А – выполнено на 33 день, Б – на 35 день пребывания в стационаре, визуализируется появление очагов пневмонии с ретикулярным рисунком в нижней правой доле с признаками уже известных кистозных поражений в нижней доле слева (белые стрелки).

treatment (piperacillin, tazobactam) and antiparasitic (albendazole) by the surgical and infectious disease services.

Discussion.

Several cases of CE have been reported in patients presenting with diverse immunosuppressive conditions, including solid cancer, chronic inflammatory disease, malignant haematological disorder, solid organ transplantation and AIDS, with development times averaging four years [7]. but there have been none to the best of our knowledge in the context of relapsed hairy cell leukaemia.

Epidemiology.

According to the World Health Organisation (WHO), E. granulosus is endemic in South America, Eastern Europe, Russia, the Middle East and China, where human incidence rates are as high as 50 per 100.000 person-years [8]. The genus Echinococcus includes six parasite species of cyclophyllid tapeworms, of which four are of public health concern and have a significant economic impact: E. granulosus (which causes cystic echinococcosis, also termed hydatid disease the genus Echinococcus includes six parasite species of cyclophyllid tapeworms, of which four are of public health concern and have a significant economic impact: E. granulosus (which causes cystic echinococcosis, also termed hydatid disease), E. multilocularis (which causes alveolar echinococcosis) as well as E. Vogeli and E. oligarthrus (which cause polycystic echinococcosis) [1]. Molecular studies have evinced this entity encompasses sheep, bovid, horse, camelid, pig and cervid strains [9, 10]. The sheep strain is the most prevalent form and is most commonly associated with human infections [9].

Most of the infections observed in Central Europe, including Germany, are diagnosed in migrants from endemic regions. Infections by imported dogs have been noted. Cystic echinococcosis in tourists visiting endemic countries is rare [1]

Life cycle.

Hosts for the cestodes tapeworm may be subdivided into three categories: definitive, intermediate, and accidental [3]. The definitive hosts are carnivores, notably dogs, cats, and wild canids, in which adult tapeworms (2.0-7.0 mm in length for Echinococcus granulosus and 1.2-4.5 mm for Echinococcus multilocularis) inhabit the small intestine [3, 11]. Domestic animals and other warm-blooded vertebrates, such as sheep, goats, cattle, horses, camels, and pigs, act as intermediate hosts, ingesting eggs released by carnivores; they carry Echinococcus granulosus predominantly. Rodents, deer, moose, reindeer, and bison are intermediate hosts of Echinococcus multilocularis [4, 12, 13]. Humans may accidentally act as an intermediate host, although

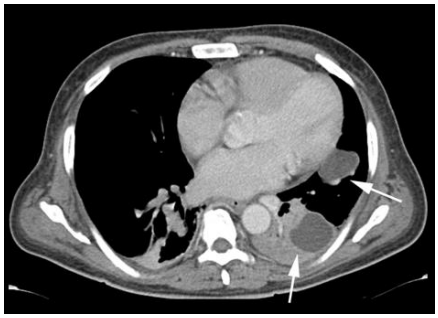
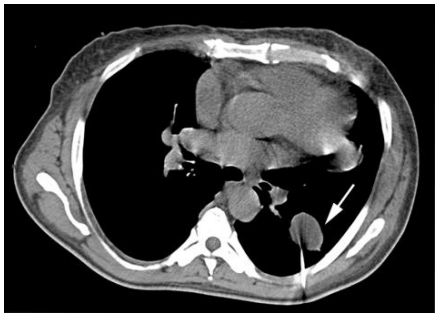


Fig. 4 a (Рис. 4 а)



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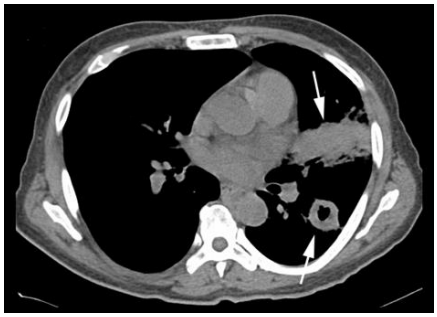

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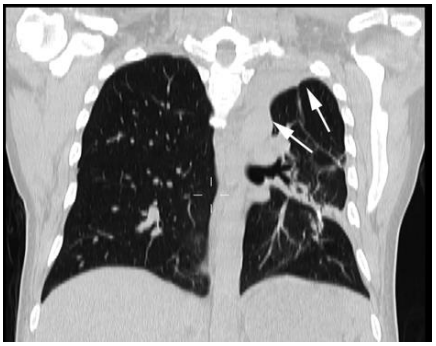

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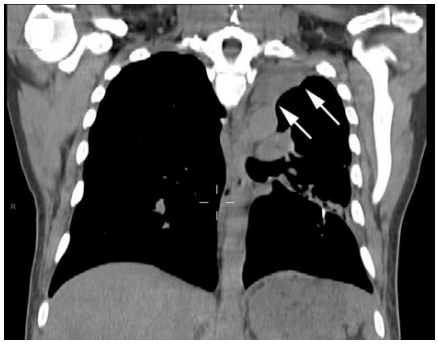

Fig. 4 e (Рис. 4 д)

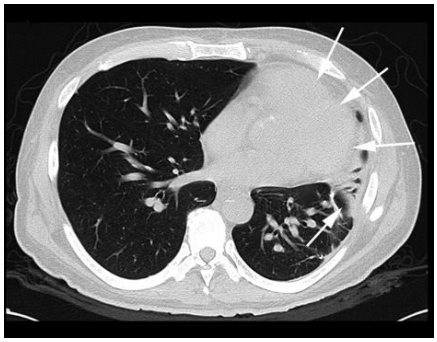

Fig. 4 f (Рис. 4 е)

Fig. 4. CT, chest.

A – an axial plane of day 35, showed the areas of pneumonic consolidation in the left upper lobe (white arrows). B – axial plane, a biopsy of a cystic lesion on day 39 confirmed the diagnosis of pulmonary hydatidosis (white arrows). C axial plane, zone of atelectasis in segment IV of the left lower lobe on day 174, and the presence of air in the already known cystic lesion (white arrows). D, E - coronal plane, resolution of the cystic lesions of the left lung on day 225, with sequelae of areas of pleural thickening in the left upper lobe (white arrows). F - axial plane, in the basal region of the left lower lobe, a zone of pleural thickening was observed, which conditioned retraction of the left anterolateral cardiac silhouette, with the presence of a residual area of pulmonary consolidation and an air bronchogram (white arrows).

Рис. 4. КТ органов грудной клетки.

А – аксиальная плоскость, на 35-й день, визуализируется консолидация легочной ткани в левой верхней доле (белые стрелки). В – аксиальная плоскость, биопсия кистозного образования на 39-й день подтвердила диагноз эхинококкоза легких (белые стрелки). С – аксиальная плоскость, зона ателектаза в IV сегменте нижней доли левого легкого на 174-й день и наличие воздуха в уже известном кистозном поражении (белые стрелки). D, E – корональная плоскость, разрешение кистозных поражений левого легкого на 225-й день, участки утолщения плевры в верхней доле левого легкого (белые стрелки). F – аксиальная плоскость, в базальной области левой нижней доли определяется зона плеврального утолщения, что обусловило западение левого переднебокового силуэта сердца, с наличием остаточной зоны легочной консолидации и воздушной бронхограммы (белые стрелки).

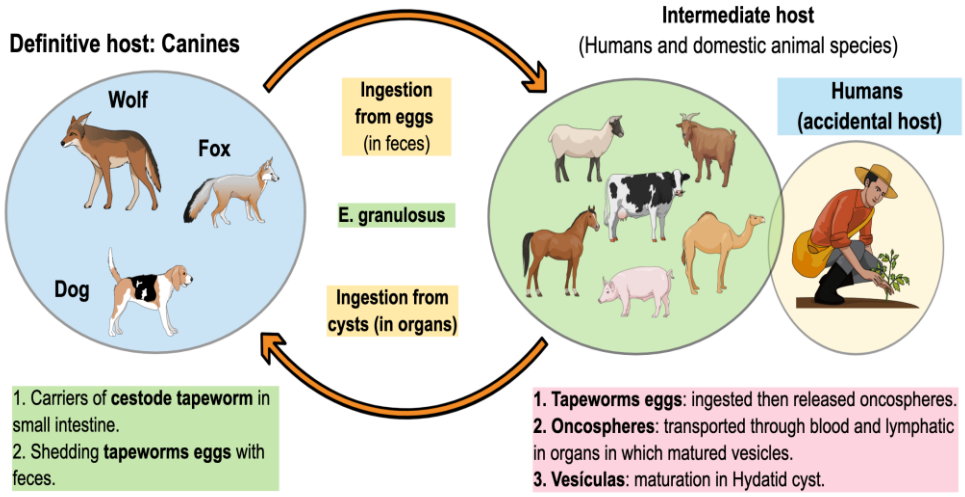

Fig. 5 (Рис. 5)

Fig. 5. Schema.

Lifecycle of Echinococcus granulosus between definitive, intermediate, and accidental hosts.

Рис. 5. Схема.

Жизненный цикл Echinococcus granulosus между окончательными, промежуточными и случайными хозяевами.

their role in the life cycle remains unclear [4, 12, 13] (Fig. 5).

A complete life cycle takes four to seven weeks. In the small intestine of definitive hosts, cestodes scolex present a double row of hooklets, which play a pivotal role in the attachment to the intestinal mucosa. Cestodes scolex have at least two proglottids (the number of proglottids changes between causative agents, from three to six in Echinococcus granulosus and two to six in Echinococcus multilocularis), which contain numerous eggs. The eggs are passed out through the host's faeces and released into the environment [3].

The intermediate hosts consume the eggs while feeding, which settle in their gut and release oncospheres later conveyed through blood or lymph to primary target organs. Lymph enters the portal circulation via the intestinal wall and travels to the visceral organs' capillary bed (primarily the liver, secondarily the lungs) [4, 12]. Oncospheres become vesicles (metacestodes) in the organs and develop concentrically into a fluid-filled cyst. Hydatid cysts for Echinococcus granulosus are initially fluid-filled, unilocular, and rich with hundreds to thousands of protoscolices; the morphology of Echinococcus multilocularis cysts differs, with masses of numerous small cysts interconnected by dense connective tissue [11]. Three layers comprise the cyst: an inner germinal and nucleated syncytial layer, surrounded by an acellular laminated layer and even further externally a host-produced fibrous adventitial layer [3, 13, 14]. Daughter cysts could grow inside larger primary cysts. Protoscolices (or protoscolex in Echinococcus multilocularis) precedes the adult worm when reuniting with the definitive host's intestine [14].

Clinical presentation.

The size of cysts in the human body is highly variable and usually ranges between 1 and 15 cm, but much larger cysts (>20 cm in diameter) may also occur [1, 4]. In humans, cysts may develop in numerous anatomic sites. This form of echinococcosis is termed primary cystic echinococcosis (CE). Secondary CE, predominantly in the abdominal and pleural cavity, results from spontaneous or trauma-induced cyst rupture and the release of protoscoleces and small cysts, which can grow to larger cysts (2, 3). Approximately 40-80% of patients with primary CE have single-organ involvement and harbour a solitary cyst. Hydatid fluid is antigenic. Thus immune-mediated reactions such as urticaria, asthma, membranous nephropathy and anaphylaxis can occur [4, 12]. Because oncospheres enter the circulation via the gastrointestinal tract, the most common site of this disease in humans is the liver

(50-70%), followed by the lungs (20-30%), and less frequently other organs including kidneys, spleen, muscles, skin, abdominal and pelvic cavities [3, 6].

Diagnosis.

Interrogation is essential to elicit recent or past long-term residence in areas of parasite transmission and work or lifestyle-related risk factors [15]. CE diagnosis in individual patients is based on identifying cyst structures by imaging techniques, predominantly ultrasonography, computed tomography, X-ray examinations and confirmation by detecting specific serum antibodies by immunodiagnostic tests [2, 3, 12].

Approximately 10-20% of patients with hepatic cysts and about 40% with pulmonary cysts do not produce detectable specific serum antibodies (IgG) and give false-negative results [3, 9].

Imaging diagnosis ordinarily results from an accidental finding in a plain chest radiograph evaluation because of the delayed growth of the cysts, becoming symptomatic with the rupture of the cysts and occasionally with the development of immune-mediated reactions against Echinococcus spp. [16-18].

As displayed in this case, the clinical diagnosis can be challenging because symptoms are unspecific [5]. Uncomplicated small cysts located in the periphery of the lung often remain asymptomatic and are detected incidentally on chest radiography [19]. Clinical symptoms occur when the cysts are large enough to exert mechanical effects on adjacent structures, and cysts larger than 5 cm in diameter can cause bronchial obstruction [20].

Complications.

Approximately 10-20% of patients with hepatic cysts and about 40% with pulmonary cysts do not produce detectable specific serum antibodies (IgG) and give false-negative results [3, 9]. Due to the slow growth of cyst development, CE can go undetected for years. Cysts can eventually exert pressure on adjacent structures depending on their size and anatomic location. They can be associated with unspecific symptoms, and when in the lungs, they might provoke a chronic cough, dyspnoea, pleuritic chest pain and haemoptysis [1].

Cysts may rupture spontaneously or due to blunt trauma and cause pleural hydatidosis with simple or tension pneumothorax and empyema or bronchial fistula. Treatment options depend on the extent of organ involvement, and the number of cysts includes both medical and surgical approaches [3].

Imaging findings.

The first step in assessing lung hydatidosis is taking a chest radiogram [8]. Uncomplicated cysts appear on chest radiographs as rounded or oval masses with smooth borders and uniform density and are surrounded by healthy lung tissue [21]. Indirect signs of lung hydatidosis on close structures (for example, trachea, bronchi) are visible if the dimensions of cysts are relevant, such as a shift of the mediastinum, pleural reactions, or compression of the lung parenchyma causing atelectasis [21, 22].

A computed tomography (CT) scan is helpful to identify better specific details of the lesions and their neighbouring structures, helping to exclude alternative differential diagnoses. In intact cysts, a CT scan may reveal a thin rim was defining the perimeter [23]. Small cysts, undetectable by a chest radiogram, may be detected with the better imaging definition provided by CT scanning, which is also valuable in the case of complex cysts; for example, it can see a cyst wall defect in a ruptured cyst [24]. Infected cysts show in CT scans as poorly defined masses with increased internal density and contrast enhancement around the cyst wall (the ring enhancement sign) after injecting a contrast substance. Computed tomography scanning can decode the cystic nature of the lung mass and supply precise localisation to plan the surgical treatment of complicated cysts [24, 25].

Ultrasonography is helpful in most cases, providing excellent images only when the cysts are nearby the pleural surface [26]. Most importantly, however, ultrasound examination of the liver may reveal concomitant liver involvement in up to 15% of individuals with lung CE [27]. Contrast-enhanced ultrasonography, based on pulsating blood flow imaging, may be used to detect small AE lesions and differentiate them from abscesses and tumours [26, 27].

Magnetic resonance imaging (MRI) might have a complementary role and theoretically better diagnostic value than a CT scan. Lung cysts exhibit hypointensity in T1-weighted images and hyperintensity in T2-weighted images, with some differences depending on the state of ripeness [21, 22]. Fluorodeoxyglucose-positron emission tomography (FDG-PET) has become the preferred reference tool for evaluating metabolic activity [28].

Differential diagnosis.

Radiologically, lung lesions can be misdiagnosed as a lung abscess caused by old tuberculosis until they are confirmed by pathological examination [29]. Also, lesions of CE can be mistaken for centrally lung carcinoma and misinterpretations of hydatid cysts for pulmonary metastases of breast cancer, respectively, have been reported [30, 31]. Vice versa, cystic pulmonary hamartomas, so-called benign metastasising leiomyoma of the uterus and bronchiectasis have been mistaken for hydatid disease [32-34].

Treatment.

For uncomplicated lung cysts, benzimidazoles are the first option; albendazole (or mebendazole) is suggested for diameters <5 cm; the cut-off size for the diameter of lung lesions is not standardised [8, 13, 35]. For that reason, the treatment usually requires the patient to undergo radical resection and long-term oral albendazole (up to 400 mg twice a day) orally; albendazole use has been reported for up to 2 years [29]. No clinical trials have compared the different treatment approaches. Albendazole can penetrate the blood-brain barrier more than mebendazole and is also the first choice of chemotherapy drugs for brain echinococcosis [36]. Cyst size, characteristics, position in the lung and clinical presentation, and the availability of medical and surgical expertise and equipment are the reasons why a consistent treatment regimen or approach may not be feasible because of the variability of pulmonary echinococcosis. Many patients with lung lesions are admitted to the hospital because of complications, mainly infection [16, 20].

In conclusion,

Cystic echinococcosis is not common in Europe but is frequently observed in the Middle East and Latin America. Diagnosis should consider a summary of the clinical, microbiological and radiological data with pathology confirmation. Imaging findings of hydatid disease can be mistaken with some benign entities, including cystic pulmonary hamartomas and bronchiectasis and malignant diseases like primary lung cancer or pulmonary metastases. Pulmonary echinococcosis should always be considered in the diagnostic work-up of pulmonary lesions in patients from endemic regions.

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