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The Case Report of the Pulmonary Larval Paragonimiasis with Simulating Cancer of the Lung

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Authors' contributions

This work was carried out in collaboration between all authors. Author VVE designed the study and wrote the protocol. Author AA wrote the draft of the manuscript. Author HAM managed the literature searches, edited and corrected the manuscript. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Larval Paragonimiasis is a parasitic disease caused by the larvae of the pulmonary fluke. In contrast to the classical Paragonimiasis, larval occurs with a large number of clinical manifestations. However, this fact only complicates the diagnosis of larval Paragonimiasis, as the abundance of clinical manifestations leads to false diagnosis of other diseases. Another distinguishing feature of this form of Paragonimiasis is the propensity to generalization and to imitation of a clinical picture of malignant neoplasms. A 66-year-old patient was infected with a larval form of Paragonimiasis, with invaded lung, pleura, heart, liver, right kidney, spleen, stomach and small omentum. Diagnostic measures performed could not give an accurate idea of the nature of the disease, so a diagnostic thoracoscopy was performed and urgent biopsy from region of dissemination was taken, which showed a glandular cancer of the lower lobe of the lung. This circumstance became the reason for performing atypical resection of the affected part, which was done, but on the second day of the postoperative period the patient died as a result of Pulmonary Embolism (PE). The cause of the disease is chronic pulmonary generalized Paragonimiasis (cyst and pneumosclerotic stages) with neoplastic syndrome.

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1. BACKGROUND

Paragonimiasis – parasitic disease of man from the class trematoda, often caused by lung Fluke *Paragonimus westermani* (the lung Fluke). Adult lung Fluke – worms are brown-red in color, they have an ovate body, convex from the back and flattened on the ventral side. Its suckers and spines which are mounted in the host organism are visible to the naked eye. Eggs Golden-brown, oval-shaped, are covered with a cap [1-2].

The rarity of this disease, but also atypical clinical picture complicates differential diagnosis and leads to a false diagnosis of other pathologies [3-6]. In medical literature, there are descriptions of cases in which paragonimiasis mimics the symptoms of tuberculosis of the lung and pleura, recurrent lung cancer or tumor metastases in the lungs [6-10]. There is evidence of cerebral form with the initial clinic hemorrhagic stroke and febrile syndrome and convulsions [11-15]. There is a description of abdominal form with the defeat of the Fluke in the pancreas, considered as a cystic-solid tumor [15].

Important diagnostic factors in paragonimiasis are: eosinophilia [1–5], positive results of ELISA and PCR [3,4], x-ray and computed tomography of the chest, where focal seals, as well as the results of PET [3,14].

As a diagnostic rarity and due to real complexities of clinical and pathological diagnosis because of atypical clinical presentation, we decided to report the case of a paragonimiasis patient who was on treatment in regional cancer centre in 2016.

2. CASE REPORT

66 years old, was admitted to the Oncology clinic with a preliminary diagnosis: Disseminated EIT of the lungs; Right-sided recurrent pleural effusion; Pulmonary insufficiency II-III degree; Ischemic heart disease; Cardiosclerosis (N-I); COPD; The tumor stage IV; Clinical group II. During admission, the patient complained of shortness of breath with minimal exertion, a feeling of heaviness and dragging pain in the right half of the chest. The patient's state was of moderate severity. Backorder thorax in the act of not breathing, rhythmic, percussion pulmonary sound, tympanic, dull in the lower divisions to the right. Auscultation: vesicular breathing weakened, in the lower left was tapped. General blood test analysis: L-8.3*109 /l, e-4.28*1012/l, HB-122 g/l, clot-203*109/l, ESR-41mm/h Biochemical blood test: ALT-183.4 u/l, total bilirubin–20 mmol/l, creatinine-132.3 mmol/l, urea 9.1 mmol/l, total protein blood-62.8 g/L. To confirm the diagnosis and determine the tactics of treatment, CT scan of the chest was conducted, which showed an encysted pleuritis of the left lung, right-sided pneumothorax with the phenomenon of compression of the lower lobe of the right lung, and pronounced fibrosis. A comparative analysis with the previous result of the CT scan, which confirmed the absence of dynamic changes in the lungs. On the basis of General examination, laboratory tests and CT of the chest, raised suspicion for the diagnosis of Cancer of the right lung. Related: pleuritis on the left; COPD; Pulmonary fibrosis; Pulmonary insuficiency II-III; Ischemic heart disease; Cardiosclerosis. Diagnostic thoracoscopy of the right and pleurodesis were performed. In the course of the operation, upon inspection of the pleural cavity the pleura was covered with fibrin, contact was bleeding. The lower lobe of the right lung was atelectic, multiple subpleural disseminates were observed. In the projection merge hollow and azygos veins were visualized in the lymph node up to 1 cm were dissected pleura, biopsy of the lymph node and urgent histological examination were diagnosed with lymphadenitis. A decision was taken to perform an atypical resection of the lower lobe of the right lung. Established in addition, thoracoport in the 5th intercostal space, atypical deleted plot lower lobes with screenings and histological examination of biopsy material were discovered isolated foci of adenocarcinoma, which were identified as glandular cancer. Under aerostatic control 10 grams of finely divided talc (pleurotes) was administered to the patient. The drainage of pleural cavity was performed at the 3 and 7 intercostal space using the Bühlau's method. The wound is sutured in layers. The next day after surgery, the patient complained only of General weakness and pain around the postoperative wound. The drainage from the pleural cavity separated light liquid. The patient was transferred from ICU to the surgical Department. At 5am on the second day after surgery, the patient suddenly lost consciousness. For resuscitation, he was urgently transferred to the intensive care unit, where he was pronounced dead.

On Postmortem examination of the corpse, in the pleura, pulmonary parenchyma, myocardium, liver, spleen, kidney, stomach, and small omentum were detected the multiple cysts of various sizes. Macroscopic signs of thromboembolism of the right branch of the pulmonary artery with hemorrhagic infarction in the right lower lobe were seen. There was noted hyperemia of the brain and internal organs. Changes characteristic of COPD were present which are as follows: thickening of the walls of the bronchi of different caliber with slimy contents, multiple foci of light patches in the parenchyma of the lungs. The thickness of the wall of the right ventricle of heart was 1.3 cm and the left ventricle wall was - 2 cm. The heart weight of 600 grams. Confirming the diagnosis of Cor pulmonale.

Histopathological examination of autopsy specimen of the brain showed perivascular edema and peri inflammatory infiltrate. In the myocardium (Fig. 1) pronounced interstitial edema with inflammatory infiltrates comprising of lymphocytes, plasma cells, eosinophils were seen. Hypertrophied muscle fibers, with multiple cysts along with the presence of the larval forms of the parasite were found in the connective tissue capsule. In the region of the pericardium were found fragments of the metacercariae of the lungs fluke with accumulations of erythrocytes (Fig. 2). In the lung tissue (Fig. 3) there were present parasitic cysts, some of which were seen to be replaced by fibrous tissue. Numbers of cysts were seen to be in close contact with the walls of the bronchi of different caliber, within the lumen were seen mucus and bronchial epithelium; interalveolar partitions were thickened. Focal lymphoadenopathy was observed around the cysts. In the liver (Fig. 4), directly under the capsule and in the parenchyma, multiple parasitic cysts were found with the presence of dead parasites. In the parenchyma of the right kidney parasitic cysts, degenerative changes of tubular epithelium, focal lymphomonocytotic infiltration were seen at the periphery of the cysts and in the interstitial spaces. In the spleen was found moderate hyperplasia of white pulp along with parasitic cysts with the presence of erythrocytes in the central part. The stomach mucosa was found to have diffuse inflammatory infiltration with erosion, with areas of fibrosis and disassociated glands and also showed presence of parasitic cysts. In the adipose tissue of the lesser omentum multiple parasitic cysts were found.

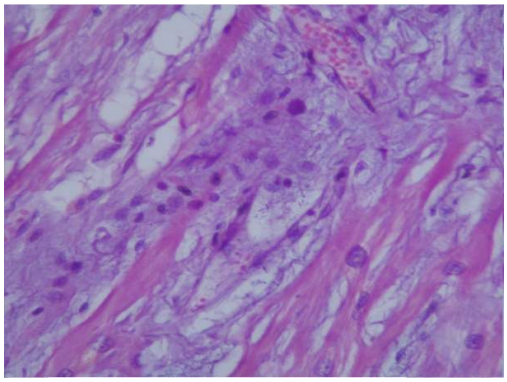

Fig. 1. Hypertrophied muscle fibers, inflammatory infiltrates containing lymphocytes, plasma cells, eosinophils. Interstitial edema. Staining with hematoxylin and eosin, x400

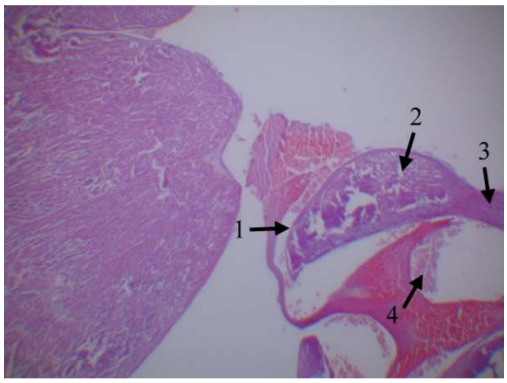

Fig. 2. The lost metacercaria of the pulmonary fluke in the pericardium: 1 - Cuticle of the larva. 2 - The body of the larva. 3 - Tail of the larva. 4 - Accumulation of erythrocytes. PAS reaction, x100

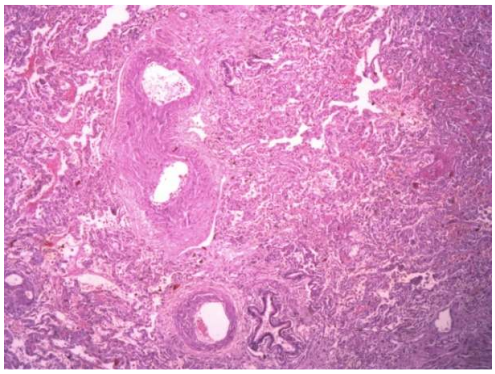

Fig. 3. Multiple parasitic cyst of the pulmonary fluke, in direct contact with the wall of the bronchioles, connected by convoluted courses. Staining with hematoxylin and eosin, x400

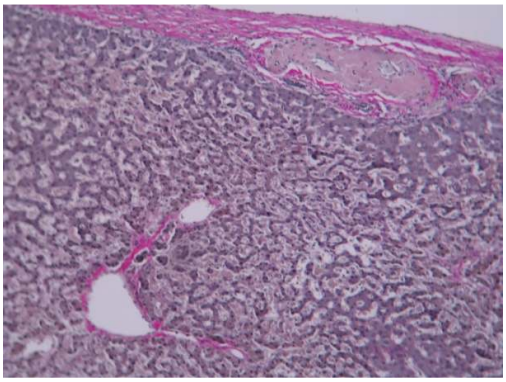

Fig. 4. Parasitic cyst in a capsule of the liver with severe fibrosis. Staining by van Gieson, x200

3. DISCUSSION

Paragonimiasis endemic regions are Asia, Africa and South America [2,3]. In the Russian Federation, it's typical for the Far East region (the Amur lower reaches and the southern coast of the Okhotsk sea) [4].

Human infection occurs most often by eating raw or insufficiently cooked, crabs, crayfish or shrimp. At the moment there are 48 species of lung Fluke, 16 of which are able to infect humans and cause paragonimiasis. All flukes can infect a person, while under metacercaria. With the help of their stiletts, they penetrate the wall of the initial section of the small intestine and into the abdominal cavity. The larvae then migrate through the diaphragm into the pleural cavity, passively invading some of the abdominal organs, particularly the pancreas, liver, lesser omentum, spleen, and stomach. After entering the pleural cavity, the larvae spend some time on the parietal pleura, and then infiltrating the viscera embedded in lung tissue, where it reaches its maturity. However, according to studies not all *Paragonimus* in the human body are able to reach sexual maturity thus remaining as larval paragonimiasis. This is because some species of the Paragonimus lung Fluke, such as *P. ichunensis, P. miyazakii, P. huatungensis* can utilise humans and some animal species as a reservoir host and, if the body is not able to respond to the invasion, Parognimiasis diagnosis becomes difficult.

In the pulmonary parenchyma the parasite develops granulomatous inflammation around itself and subsequent fibrotic encapsulation with the formation of cysts and the possible penetration of the worms into the lumen of the bronchioles. Cysts may reach 2 cm in diameter. Their number can vary depending on the severity of the infectious process.

Patients with paragonimiasis reveal a wide variety of radiographic and CT findings, including pulmonary nodules or masses, and sometimes they are suspected to have a lung tumor [16]. FDG (fluorodeoxyglucose)-PET (positron emission tomography) is used recently to differentiate benign from malignant lesions, and it has been shown to be more effective than CT [17]. However, FDG is not a cancer-specific agent, and false positive findings in benign diseases, including paragonimiasis, have been reported [18-21]. Earlier studies have reported that the radiologic findings of pulmonary paragonimiasis included pleural effusion, hydropneumothorax, pulmonary nodules, or air-space consolidation, and cysts [22].

The diagnosis of paragonimiasis can be made by detecting eggs in the tissue section, or more commonly, in the stool, sputum, or BAL fluid, or by a positive anti-*Paragonimus* antibody test. However, egg detection rates have been reported to be low (28-38%) and eggs are not present in the sputum until 2-3 months after an infection. Serum determination of antibodies to *Paragonimus* is more accurate in early infections, and ELISA is highly sensitive (92%) and specific [15].

4. CONCLUSIONS

This case report highlights the complexity of the diagnosis of cystic pneumosclerotic phase of generalized larval paragonimiasis occurring with tumor syndrome. The feature of monitoring is that the patient had disseminated paragonimiasis with multiple organ lesions, fibrogenetic formation of multiple cysts in the affected organs. Poor diagnosis and post operative management of the case contributed to the death of the patient. Paragonimiasis was not diagnosed in a timely manner, specific treatment was not conducted, which contributed to the progression of the inflammatory process in the lungs, with involvement of many organs, with the development of pulmonary heart disease, pulmonary embolism, which was the immediate cause of death. The discrepancy between diagnoses is due to a rare pathology with an atypical clinical presentation (Category II).

CONSENT

As per international standard or university standard, patient's written consent has been collected and preserved by the authors.

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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