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Case Report

Laparoscopic excision for ectopic peritoneal paragonimiasis mimicking a gastric duplication cyst: A case report

Chul Kyu Roh [a], Min Jung Jung [b,*]

[a] Department of Surgery, Ajou University School of Medicine, Suwon, South Korea
[b] Department of Pathology, College of Medicine, The Catholic University of Korea, Seoul, South Korea

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ABSTRACT

Introduction: Paragonimiasis, lung fluke disease caused by infection with *Paragonimus* species, is a food-borne parasitic zoonosis. The overriding symptoms of *Paragonimus westermani* infection include chronic cough, shortness of breath, and pleuritic pain. Extrapulmonary paragonimiasis caused by aberrant parasitic migration is known to occur in a variety of sites such as the brain, abdominal wall, and intraperitoneal cavity. Ectopic paragonimiasis is an uncommon disease that presents with a few clinical manifestations, which makes it difficult to diagnose and treat.

Case presentation: A 47-year-old man with an unremarkable medical and surgical history presented with a peritoneal lesion that was discovered incidentally on abdominal computed tomography during routine health screening. The patient did not exhibit any associated symptoms such as abdominal pain. The radiologic diagnosis was a gastric duplication cyst and we performed laparoscopic excision of the peritoneal mass. Histopathological examination revealed paragonimiasis, and the result of the skin test for paragonimiasis was positive. The patient was treated with praziquantel.

Clinical discussion: The diagnosis of ectopic peritoneal paragonimiasis remains challenging due to inexperience, misdiagnosis, and its rarity. Clinicians should bear in mind that an intra-abdominal mass may be related to a parasitic infection. The detection of the ova of *Paragonimus* parasites in sputum and biopsy specimens may be difficult due to an insufficient amount.

Conclusion: Clinicians need to thoroughly take the patient's history and clinically suspect parasitic infections. Laparoscopic resection of this rare mass is safe, feasible, and allows for rapid recovery.

1. Introduction

Paragonimiasis is a parasitic zoonosis caused by various species of *Paragonimus*. More than 10 species have been reported to cause human infections with the commonnest being the oriental lung fluke, *Paragonimus westermani.* [1] Humans are infected via ingestion of raw, partially cooked, or pickled crustaceans [2]. Infections of the genus *Paragonimus* occur in various parts of the world, particularly South America, West Africa, and East Asia [1,3]. In 2012, approximately 290 million individuals worldwide were at risk of paragonimiasis, and it was reported that approximately 20 million individuals were infected worldwied [4].

Paragonimiasis presents with pulmonary and extrapulmonary manifestations. The common pulmonary manifestations include chronic cough, hemoptysis, and pleuritic chest pain [5,6]. Extrapulmonary manifestations have been reported in the central nervous system, pericardium, mediastinum, peritoneal cavity, genitourinary system, and subcutaneous tissues [7–10]. The brain is the most frequent site of extrapulmonary paragonimiasis [11].

In recent years, advances in hygienic practices and changes in food consumption patterns have reduced the incidences of parasitic diseases. Paragonimiasis has become an overlooked disease in Korea as the number of patients with the disease declined [12,13]. Furthermore, the rarity of abdominal paragonimiasis make it difficult for clinician to diagnose since it may be confounded with other abdominal diseases such as diverticulitis, abscess, tumor, and carcinomatosis [14–18]. Herein, we present a case of ectopic peritoneal paragonimiasis mimicking a gastric duplication cyst, which was successfully resected via the laparoscopic

* Corresponding author. Department of Pathology, Bucheon St. Mary's Hospital, College of Medicine, The Catholic University of Korea, 327, Sosa-ro, Wonmi-gu, Bucheon-si, Gyunggi-do, 14647, Republic of Korea.
E-mail address: ajmania@naver.com (M.J. Jung).

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approach. This report also highlights the importance of this approach in the treatment of pathologies of the abdomen. This case report was written according to the Surgical Case Report guidelines (SCARE) [19]. This case report was approved by our Institutional Review Board (Approval No. AJIRB-MED-EXP-21-270), which waived the requirement for informed consent because of full anonymization of patient information.

2. Presentation of case

A 47-year-old man was referred to our outpatient clinic for an intra-abdominal lesion observed on chest computed tomography during a routine health screening. The patient had no past medical, surgical, family, psychological and pharmacological histories. He did not complain of any abdominal symptoms related to the lesion. He did not have any history of surgeries, history of allergies, or adverse drug reactions. Physical examination revealed no gross abnormality or localized tenderness in the abdomen.

Results of initial laboratory tests, including complete blood count and blood chemistry panel, were as follows: White blood cell count and absolute eosinophil count were 3400/μL and 95/μL (reference range, 0–500/μL), respectively. The eosinophil percentage was 2.8% (reference range, 0–10%). The hemoglobin level and platelet count were 13.8 g/dL and 239,000/μL, respectively. The results of other routine biochemical analyses, including blood urea nitrogen, creatinine, proteins, albumin, liver enzymes, amylase, lipase, prothrombin time, and partial thromboplastin time were within the normal limits.

His chest radiograph was normal. Abdominal computed tomography revealed a mass-like lesion abutting the stomach, which was suspected of being continuous with the gastric mucosa (Fig. 1A). It appeared slightly heterogeneous with low attenuation without significant enhancement. The size of the lesion was estimated to be approximately 2.9 × 2.3 cm [2] (Fig. 1B). Abnormal lymphadenopathy was not observed in the peritoneal and retroperitoneal spaces. The radiological diagnosis was gastric duplication cyst, and the differential diagnosis included gastric subepithelial tumor, pancreatic cystic tumor, pseudocyst of spleen., and mesenteric cyst. Esophagogastroduodenoscopy, which was performed for routine evaluation, revealed chronic gastritis without any pathological lesion.

Dr. CK Roh performed laparoscopic surgery for resection of the intra-abdominal mass. After induction of pneumoperitoneum with carbon dioxide gas, we approached the mass through the lesser sac. The mass was located in the upper body and posterior wall of the stomach; however, it did not arise from the stomach lining. We meticulously dissected the area around the mass using a laparoscopic energy device, and the mass was excised without breaching the capsule. The mass did not originate from the adjacent organs, such as the stomach, pancreas, and spleen. We removed the mass from the abdominal cavity within the endoscopic bag to avoid surgical site contamination. The operation took approximately 30 minutes from skin incision to wound closure. There were no intraoperative complications such as blood transfusion and unplanned conversion. Surgery was successful and the patient was discharged on postoperative day 1 without any complications.

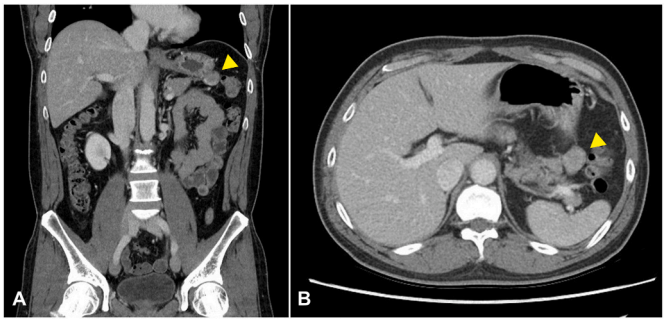

Fig. 1. Abdominal computed tomography image during routine health screening. (A) Coronal view shows a mass-like lesion abutting the stomach, which was suspected to be continuous with the gastric mucosa(yellow arrow). (B) Axial view shows slightly heterogeneous mass with low attenuation without significant enhancement (yellow arrow). . (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

Histopathological assessment revealed that the gray cystic mass measured 4 × 2 × 2 cm^3. The cut surface had a unilocular cystic appearance, with dirty yellow material within it (Fig. 2). The microscopic section revealed fibrocystic structures with a few parasitic eggs in the wall of mass. Several degenerative eggs were observed in the necrotizing granuloma. Old semi-degenerated parasites were also identified along with extensive fibrosis and mild inflammatory reaction around them. These features were consistent with those of paragonimiasis with degeneration.

After the diagnosis of paragonimiasis, the patient underwent parasite-specific antibody testing. The result of the *Paragonimus westermani* antibody test was positive (optical density value, 2.43), while the results of antibody tests for *Cysticercus*, *Sparganum*, and *Clonorchis sinensis* were negative (optical density value < 1.0). Thereafter, detailed patient history was obtained. The patient had eaten soybean sauced freshwater crabs several times. The patient was orally administered praziquantel for treatment (25 mg/kg, 3 times a day for 3 days) after establishing the diagnosis of peritoneal paragonimiasis. The patient was followed up for more than 6 months. Recent the white blood cell count and normal eosinophil count were within the normal range. The patient had not complained of any symptoms.

3. Discussion

Previous studies have reported laparoscopic approach of peritoneal paragonimiasis identified in falciform ligament, right colon, and gastric wall [17,20,21]. Here, we report on laparoscopic excision for ectopic peritoneal paragonimiasis mimicking gastric duplication cyst. With the recent improvements in laparoscopic techniques and the development of laparoscopic instruments, laparoscopic approaches may be particularly beneficial for benign abdominal pathologies.

Humans are usually infected by ingestion of uncooked crustacean such as a crab or crayfish that harbor the encysted metacercariae of the *Paragonimus* species. The metacercariae excyst in the duodenum, penetrate through the intestinal wall, and enter the peritoneal cavity. Additionally, they can pass through the diaphragm into the thoracic cavity and reach the lung parenchyma where they mature into adult flukes (7–12 mm in length). These adult flukes are found encapsulated in the lungs [1,11]. The metacercariae can reach and develop ectopic lesions anywhere in the body through this route of migration from the intestine to the lungs [11]. On rare occasions, the adult flukes may be found encapsulated in the brain or peritoneal cavity [8,22]. The most common extrapulmonary infections are observed in the brain and peritoneal

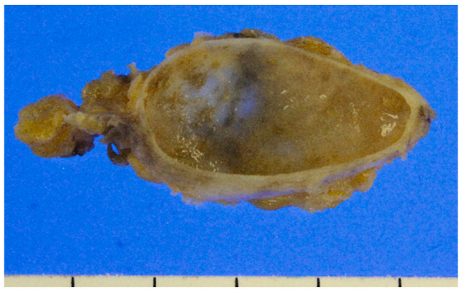

Fig. 2. Photograph of the gross specimen (cross section). The gray unilocular cystic mass contained dirty yellow material. . (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

infections occur mostly in the liver and colon [11,23].

Due to its rarity, paragonimiasis is considerably difficult to diagnose, and the appropriate examinations are often neglected. Furthermore, suspecting this parasitic infection is not easy before obtaining histopathological confirmation in cases of extrapulmonary paragonimiasis. Our patient had no subjective symptoms, and none of the abnormal laboratory results suggested a parasitic infection. Thus, we did not suspect a parasitic infection as a differential diagnosis of this patient's intra-abdominal mass.

High levels of eosinophils in the peripheral blood and high serum immunoglobulin E levels are helpful in establishing the diagnosis, although they are non-specific findings of parasitic infection. This patient did not present peripheral blood eosinophilia. Therefore, paragonimiasis was diagnosed according to histopathological evaluation of the mass that was laparoscopically excised and serologic test for *Paragonimus*-specific immunoglobulin G antibody. The intradermal reaction to the *Paragonimus* antigen is the most common test used to screen for paragonimiasis. However, antibody test should be judged comprehensively in clinical practice since antibody levels remain high for years even after effective treatment.

With initial diagnosis, gastric duplication cyst is rare congenital anomaly, and standard treatment is complete surgical resection due to the risk of malignant transformation. Gastric duplication cyst is definitely confirmed after pathological assessment. Based on the initial diagnosis, the patient underwent laparoscopic surgery and did not require additional analgesics for postoperative pain, recovered rapidly, and quickly returned to normal activity. We recommend the laparoscopic approach as a treatment option for abdominal masses, especially benign or non-malignant lesions, including parasitic neoplasms.

4. Conclusion

Ectopic paragonimiasis is difficult to diagnose since it is unfamiliar and rare. In this patient, the diagnosis was possible only after histopathological examination of the cystic mass resected via laparoscopic surgery, which emphasizes the importance of comprehensive clinical history taking and high clinical suspicion of parasitic infections. Clinician should include ectopic paragonimiasis in the differential diagnosis of intra-abdominal masses. The laparoscopic approach appears to be the standard treatment option for abdominal masses, especially benign or non-malignant lesions, including parasitic neoplasms.

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Ethical approval

The Institutional Review Board of Ajou University Hospital, Suwon, Korea, approved this study (approval number: AJIRB- MED-EXP-21-270).

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Chul Kyu Roh: Conceptualization, Investigation, Writing – original draft. Min Jung Jung: Investigation, Conceptualization, Writing – original draft, Writing – review & editing.

Guarantor

Min Jung Jung, MD.

Provenance and peer review

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Declaration of competing interest

The authors have no potential conflicts of interest to disclose.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.102754.

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