



Case Report

Incidental diagnosis of paragonimiasis after histopathological examination of cholecystectomy specimen in Nepal: A case report

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ABSTRACT

Introduction: Paragonimiasis or lung fluke disease is a typical food-borne parasitic zoonosis caused by infection with *Paragonimus species*. The *Paragonimus* is a trematode that mainly infects the lungs of humans after eating an infected raw or undercooked crab or crayfish.

Case presentation: Herein we report a case of peritoneal Paragonimiasis in the gallbladder of a 58-year-old female from Rukum district of Nepal. It was an incidental diagnosis following routine histopathological examination of the cholecystectomy specimen. She presented with the symptoms of abdominal pain, fever, cough, and had a history of consumption of partially cooked river crabs. She responded well to praziquantel and improved thereafter.

Clinical discussion: Ectopic paragonimiasis is a rare disease and it presents with few clinical symptoms so it is significantly difficult to make a diagnosis and treat the patients. Even if a sputum test and biopsy are performed, the ova or body of *Paragonimus* parasites may not be detectable due to insufficient amount of specimens. Therefore, thorough history taking should be given importance. The dietary history of partially cooked crab or crayfish should indicate towards a high suspicion of Paragonimiasis. Any such indication should be immediately confirmed, which in our case was done by routine histopathological examination of the cholecystectomy specimen.

Conclusion: We report the case of a patient with peritoneal paragonimiasis in the gallbladder. Ectopic paragonimiasis is hard to diagnose due to an ignorance of, misdiagnosis, and the rarity of this disease. Thus, thorough history-taking and clinical suspicion of parasitic infection is essential.

1. Introduction

Paragonimiasis is a parasitic zoonosis caused by infection with *Paragonimus* species. It typically is a food borne disease and occurs by eating raw, smoked, partially cooked crustaceans (crayfish or crabs). Lung flukes are present in various organs other than the lungs, such as the brain, peritoneum, subcutaneous tissue, and retroperitoneum. Owing to its rare existence, abdominal paragonimiasis poses a major diagnostic problem for clinicians and can be confused with other abdominal diseases [1]. This case report has been reported in line with the SCARE 2020 criteria [2].

Herein we report a case of peritoneal Paragonimiasis in the gallbladder. It was an incidental diagnosis following routine histopathological examination of the cholecystectomy specimen. Therefore, this

study highlights the importance of routine histopathology of gallbladder after elective cholecystectomy.

2. Presentation of case

A 58-year-old female from the Rukum district of Nepal with complaints of abdominal pain, fever, and cough was brought to the outpatient department by her husband via public vehicle. History of her dietary habits revealed frequent consumption of smoked and improperly cooked crabs collected from the mountain streams in Rukum. There was no history of pleural effusion, ascites and tuberculosis. No history of tuberculosis in the family. No surgical history in the past. No history of allergy or adverse drug reaction. Patient was treated with Praziquantel for peritoneal paragonimiasis, apart from that there was no significant

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treatment history. Her complete blood count (CBC) revealed a total leukocyte count of 17,000 cells/mm³ with 59% eosinophils. It also showed an increased erythrocyte sedimentation rate (ESR). The rest of the blood investigations were within normal limits. Her ultrasonography (USG) showed cholelithiasis for which cholecystectomy was done subsequently after three days. Routine histopathology of the gallbladder after elective cholecystectomy was sent. It was done in a private hospital in Nepal by a senior consultant pathologist. Macroscopically, there were multiple small round, smooth-surfaced nodules throughout the gallbladder wall. The gallbladder measured 5.6cm * 3.5 cm * 2.9 cm. An incidental diagnosis of Paragonimiasis after demonstration of various *Paragonimus* parasites in the gall bladder tissue was made. The microscopic section showed old semi degenerate parasites, with extensive fibrosis of the gallbladder wall (Fig. 1a), mild inflammatory reaction around the parasites (Fig. 1b) along with chronic follicular cholecystitis (Fig. 1c).

The confirmed diagnosis of Paragonimiasis was based on the presence of *Paragonimus* parasites in the gallbladder tissue as well as high absolute eosinophilia in her peripheral blood. She was treated with 25mg/kg of Praziquantel three times a day for three days, responded well to treatment, and was recovered fully on follow-up after 8 weeks.

3. Discussion

Paragonimiasis is a zoonosis caused by many species of *Paragonimus* commonly *P. westermani*. Humans are infected by eating raw, smoked, or partially cooked crustaceans (crayfish or crabs) [3]. Among food-borne trematodes, the lung fluke infection costs more in terms of disability-adjusted life years (DALYs) than opisthorchiasis, fascioliasis, and intestinal diastole infection combined as they are usually misdiagnosed as tuberculosis, pneumonia, or pleural effusion. An estimated 293 million people are at risk of paragonimiasis worldwide and it has been reported that about 23 million people in 48 countries are infected [3,4].

Humans are usually infected when they consume inadequately cooked crabs or crayfish, which contain the encysted metacercariae of *Paragonimus* species. The metacercariae can pass through the intestinal wall into the peritoneal cavity and through the diaphragm into the pleural cavity, eventually ending up in the lung parenchyma and finally

growing into adult flukes [5]. Because of this migratory route from the intestine to the lungs, the worms may reach and develop in ectopic foci anywhere within the body, including the liver, gallbladder, spleen, intestines, subcutaneous tissues, or brain. The brain is the most common site of extrapulmonary involvement [6]. On the other hand, peritoneal infections are not common and occur mostly in the liver and colon [7,8]. No report has been published regarding the diagnosis of paragonimiasis in the gallbladder, with the help of cholecystectomy histopathological specimen according to our understanding. Although the diagnosis of ectopic paragonimiasis via histopathological specimen of the brain, liver, spleen, abdominal wall, orbital cavity, skin, and omentum have been reported. Routine histopathology of the gallbladder after elective cholecystectomy has helped in the incidental diagnosis of chronic cholecystitis alone, acute cholecystitis with mucocele, acute cholecystitis with empyema, chronic cholecystitis associated with polyp and adenocarcinoma of varying differentiation along with cholelithiasis [9]. This shows the importance of routine histopathology of the gallbladder after elective cholecystectomy.

Major clinical symptoms include chest pain, difficulty in breathing, and rusty brown cough or blood-stained stool, or recurrent hemoptysis. However, in our case, only cough is seen with fever and abdominal pain. There is no difficulty in breathing or any stain seen in the blood or cough.

Due to the rarity of the disease, it is significantly difficult to make a diagnosis of Paragonimiasis and proper examinations are often not performed [10]. A high eosinophil count in peripheral blood and an elevated serum IgE values are not specific findings; they are observed in around 80% of patients [11]. In this case, the patient showed peripheral blood eosinophilia. Even if a sputum test and biopsy are performed, the ova or body of *Paragonimus* parasites may not be detectable due to an insufficient amount of specimens [12]. However, in our case, the microscopic section of the gallbladder tissue showed old semi degenerate parasites, surrounded by extensive fibrosis and calcification. A similar study done by Lee et al. has suggested that the larvae probably matures in the peritoneal cavity into adult flukes that later die, leaving nodules of fibrous tissue, calcification, and eggs [13].

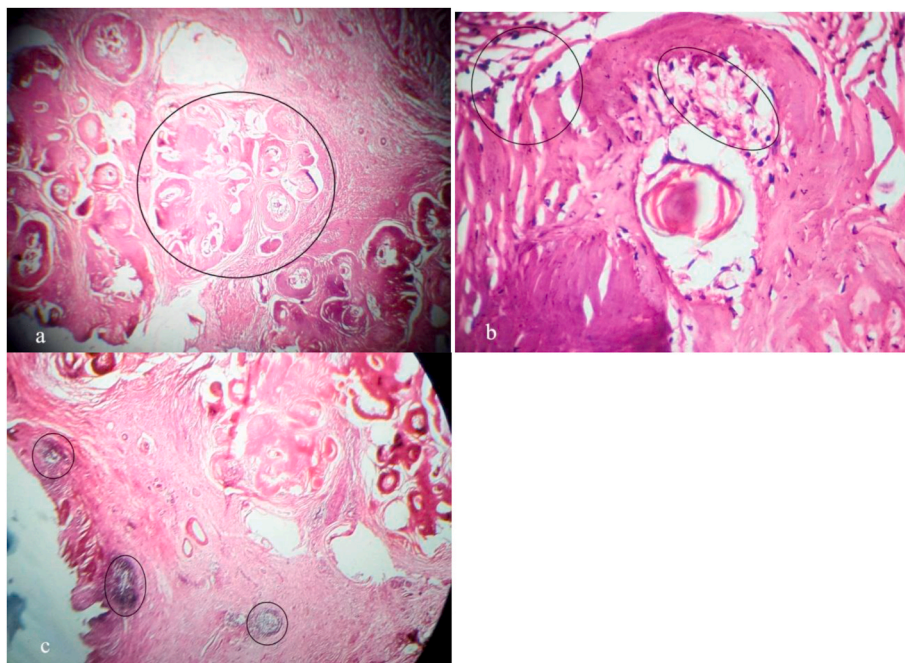


Fig. 1. Microscopic findings of Paragonimiasis in cholecystectomy specimen

4. Conclusion

Paragonimiasis is a lung fluke that can also be present in ectopic sites such as the brain and peritoneum. We report the case of a patient with Peritoneal Paragonimiasis in the gallbladder. It was an incidental diagnosis following routine histopathological examination of the cholecystectomy specimen. Therefore, the histopathological examination must be carried out in all cholecystectomy specimens for incidental diagnosis of various diseases. In addition, ectopic paragonimiasis is hard to diagnose due to an ignorance of, misdiagnosis, and the rarity of this disease. Thus, thorough history-taking and clinical suspicion of parasitic infection are essential.

Author agreement statement

We the undersigned declare that this manuscript is original, has not been published before and is not currently being considered for publication elsewhere.

We confirm that the manuscript has been read and approved by all named authors and that there are no other persons who satisfied the criteria for authorship but are not listed. We further confirm that the order of authors listed in the manuscript has been approved by all of us.

We understand that the Corresponding Author is the sole contact for the Editorial process. He/she is responsible for communicating with the other authors about progress, submissions of revisions and final approval of proofs.

Ethical approval

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Authors contributions

Author 1: Led data collection, contributed to writing the case information and discussion.

Author 2: Contributed to the process of original draft preparation and introduction.

Author 3: Contributed to conceptualization, methodology, and discussion.

Author 4: Revised it critically for important intellectual content, contributed to review and editing.

Author 5: Edited the rough draft into the final manuscript.

Author 6: The pathologist, who diagnosed the case, collected the data, and preserved the pictures.

Author 7: The microbiologist, who helped in the diagnosis and supervised throughout the process of manuscript writing.

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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.

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Declaration of competing interest

None.

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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.2021.02.016>.

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