Case Report

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Diagnosis of a pulmonary hydatid cyst by fine needle aspiration: a case report with literature review

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Introduction and importance: Hydatid cysts are seldom identified in cytologic smears and are often incidental. This report highlights a case where fine needle aspiration cytology was unintentionally utilized to diagnose a pulmonary hydatid cyst clinically mistaken for an abscess.

Case presentation: A 29-year-old female presented with intermittent respiratory symptoms, including a mild cough and sputum, that she has been complaining of since 2020. A blood investigation revealed an elevated erythrocyte sedimentation rate (25 mm/h), C-reactive protein (> 5 mg/dl), and white blood cells ($> 11 \times 10^9$/l). A high-resolution computed tomography scan of the chest revealed an irregular pulmonary opacity in the right lower lobe with central fluid attenuation, suspecting an abscess or, less likely, a pulmonary neoplasm. The patient underwent fine needle aspiration cytology, which was diagnosed as a hydatid cyst. A thoracotomy was performed, and the cyst was totally excised. Histopathological results of the specimen confirmed the previous diagnosis.

Clinical discussion: Various diagnostic modalities have been discussed for diagnosing hydatid cysts, including radiology, serology tests, and fine needle aspiration. These modalities have been evaluated in terms of their respective advantages and drawbacks.

Conclusion: Hydatid cysts can be challenging to diagnose based on imaging findings and may be misidentified as abscesses or masses. Despite the risk of anaphylaxis, fine needle aspiration cytology can be a reliable diagnostic method if performed meticulously by experienced radiologists.

Keywords: diagnostic modalities, echinococcosis, FNAC, hydatid disease, parasitic infection

Introduction

Hydatid disease is a widely recognized zoonotic-parasitic infection. It is more common in the developing countries[1,2]. It is caused by the larval stage of *Echinococcus granulosus*. Humans typically serve as intermediate hosts, becoming infected either through direct contact with the primary hosts or by consuming contaminated food and water with the parasite's eggs[2,3]. *E. granulosus* and *E. multilocularis* are the two familiar species of the echinococcus parasite responsible for causing cystic echinococcosis and alveolar echinococcosis in humans. The liver (70%) and lungs (20%) are the most frequently affected organs[4]. Upon ingestion of the parasite's eggs by intermediate hosts, the hatched embryos traverse the intestinal wall and enter the portal circulation, initially affecting the liver. A few embryos may have the ability to migrate through the hepatic veins and reach the lungs[3]. The diagnosis of the disease is generally complicated due to varied clinical presentations or nonspecific symptoms. The disease can remain silent or asymptomatic for years until the cyst grows large enough to cause compression and trigger symptoms[2]. Although fine needle aspiration cytology (FNAC) is useful for diagnosing various inflammatory, fungal, and parasitic diseases, echinococcosis is seldom identified in cytologic specimens and is often incidental. Furthermore, the cytological features of hydatid cysts (HCs) are not well-established and can be misdiagnosed with cysticercosis[5]. Notwithstanding these constraints, a few studies have reported the diagnosis of hydatid disease through FNAC[5–10]. This report highlights a case where FNAC was unintentionally utilized to diagnose a pulmonary HC initially mistaken for an abscess. The report has been written according to the SCARE guideline and the references have been confirmed to be eligible[11,12].

HIGHLIGHTS

- Diagnosis of the disease is frequently complicated due to varied clinical presentations or nonspecific symptoms.
- Fine needle aspiration cytology is useful for diagnosing various inflammatory, fungal, and parasitic diseases.
- Echinococcosis is seldom identified in cytologic specimens and is often incidental.

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Case presentation

Patient information

A 29-year-old female presented with intermittent respiratory symptoms, including a mild cough and sputum, that she has been complaining of since 2020. During the COVID-19 outbreak, the patient developed a fever and body pains for about ten days and was diagnosed with a COVID-19 infection. She received Panadol Extra (1 mg/day) for 5 days, and her condition recovered after 10 days of isolation, but she later developed intermittent respiratory symptoms like a mild cough and sputum. At that time, she was treated by an internist with periodic antibiotics (amoxiclav tab, 1 gm, 1×2) and steroids for 7 days which resulted in symptomatic improvement. Recently, the patient's respiratory symptoms started to bother her again.

Clinical findings

On the physical examination, she had only mild respiratory symptoms as mentioned, and no other significant findings were noticed. A blood investigation revealed elevated inflammatory markers like erythrocyte sedimentation rate (ESR) (25 mm/h), C-reactive protein (CRP) (>5 mg/dl), and white blood cells (WBC) ($>11 \times 10^9$/l).

Diagnostic assessment

Due to the persistence of the patient's symptoms, a high-resolution computed tomography (HR-CT) scan of the chest was performed and revealed an irregular pulmonary opacity in the right lower lobe with central fluid attenuation (Fig. 1). It was suspected to be an abscess or, less likely, a pulmonary neoplasm.

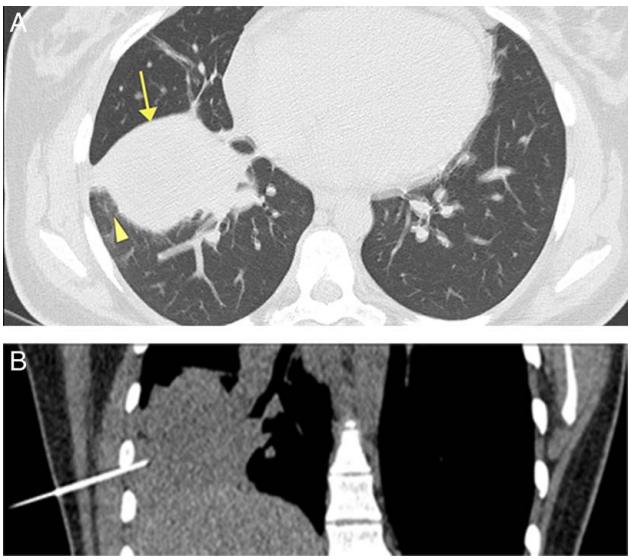

Figure 1. (A) Axial non-enhanced computed tomography (CT) (pulmonary window) showed a large pulmonary opacity "arrow" at the right lower lobes which is associated with subtle surrounding pulmonary inflammatory change "arrowhead" on the form of surrounding ground glass opacity. (B) Selected coronal CT image showed pulmonary lesion showed apparent central fluid attenuation, the fine needle aspiration needle tip is noted within the presumed wall of the cavitary lesion (not centre).

Therapeutic intervention

The patient underwent an FNAC on the right hemithorax in a prone position, which suggested the presence of an HC (Fig. 2). Thereafter, a thoracotomy was performed, and the abscess was removed. Histopathological results confirmed the previous diagnosis.

Follow-up

The postoperative period was uneventful, and the patient's symptoms improved.

Discussion

Hydatidosis is caused by the immature stage of E. granulosus, in which the adult form usually resides in the intestines of dogs and other canines. Humans, as intermediate hosts, are commonly affected during childhood because of their close contact with pets. However, HCs take a long time to develop and induce symptoms, resulting in frequent diagnoses in middle-aged or elderly individuals[5,8]. Typically, patients with abdominal HCs do not experience symptoms unless the cysts become complicated by enlarging size, rupture, or infection. Asymptomatic cases are often diagnosed accidentally during routine radiological exams for other illnesses. In some cases, it may present with nonspecific symptoms, such as the presence of a mass or pain[2,13]. The present case was a young female patient who complained of a mild cough and sputum for two years.

Regarding the diagnostic modalities for diagnosing HCs, different assumptions have been discussed[5,7]. The accuracy of radiology in diagnosing HCs is not universally reliable. Computed tomography (CT) or MRI scans may depict lesions with diverse appearances, such as isodense, hyperdense, homogeneous, or heterogeneous. Consequently, this may lead to misdiagnosis as an abscess or a neoplastic condition, as encountered in the present case. The limited accessibility and high cost of MRI make it less suitable for rural areas in developing countries[5]. Although, others believe that radiological investigations like

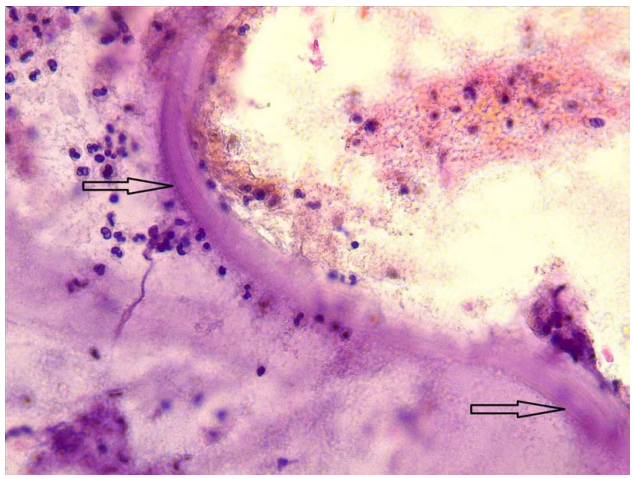

Figure 2. The aspirate revealed cellular eosinophilic material containing a laminated membrane (black arrow) of hydatid cyst with inflammatory cell infiltrate.

ultrasound (U/S), CT, and MRI are useful for diagnosing hydatid disease by demonstrating the characteristic appearance of cysts, their size, localization, and relationship to adjacent tissues. They can also help identify the disease in other locations[7,14]. Despite often yielding positive results, serology can produce false negatives when senescent or deceased cysts are present[5]. In the diagnosis of HC by serology, Rickard *et al*.[15] found a significant false-negative rate of 20%. Serological tests show limited sensitivity and specificity for diagnosis. These tests may be complementary to pathological and radiological examinations. If the features of an HC are detected through cytology and radiology, serological tests may not be necessary[7].

Cytologic techniques have been endorsed for diagnosing palpable lesions, but performing and interpreting aspirations require significant skill and manual dexterity. It is generally acknowledged that this method possesses a high specificity and sensitivity. Although variations in accuracy can arise based on the location and characteristics of the lesion[5]. FNAC is a well-established and reliable method for diagnosing HCs in the liver. While due to the potential risks of anaphylaxis and cyst spread, performing FNAC in suspected HC cases is considered a high-risk procedure. In Von Sinner and colleagues' study of 31 FNA cases for diagnosing HCs, unintentional use of the tool occurred in 18 cases, while intentional FNA was performed in 13 cases. Among the patients, 25 had no reactions, while five experienced minor allergic reactions. Furthermore, one patient had a sudden severe drop in blood pressure, necessitating antishock therapy, but eventually recovered without complications[16]. Das and colleagues reported that out of eight cases, accurate diagnosis of HC through FNAC was achieved in only five cases. In contrast, a study by Sinan and colleagues involving 55 cases demonstrated that 44 cases were accurately diagnosed using the U/S, and in the remaining 11 cases, U/S-guided FNAC successfully provided a definitive diagnosis[13,17]. The FNA sites of the 11 cases reported by Sinan *et al*.[13] included the liver in 7 cases and one case per kidney, abdominal wall, retroperitoneum, and neck. In the cases reported by Das *et al*.[17], the FNA sites were the liver in six cases and one case per lung and mediastinum. Because the MRI findings in the present case did not suspect HC but rather an abscess or a neoplasm, the patient underwent an FNAC. The result of the FNAC came back as HC without any complications due to the procedure. However, performing FNAC in suspected cases is considered a high-risk procedure. In our case, the success of the procedure might be attributed to the experience of the radiologist.

Cytology findings in HC can vary depending on the aspiration site and cyst duration. Smears from HC typically exhibit the morphology of the three layers. The most frequently observed element in the smear is the laminated cyst wall. Moreover, protoscolices, a germinal layer, and hooklets can also be detected[18]. A study proposed that only the presence of irregular fragments suggestive of a laminated layer is sufficient to consider HC as a definitive diagnosis[6]. In contrast to the previous study, other authors reported that only hooklets and granular debris could be seen in the cytologic smear and defined scolices and hooklets as indicators of HCs at FNAC[19]. In the study conducted by Das and colleagues, the cytological smears of eight HC cases showed various combinations of features. One case showed a laminated cyst wall, scolices, and hooklets. Two cases exhibited scolices and hooklets, and two other cases showed a laminated cyst wall along with hooklets. The last three cases presented only the laminated cyst wall[16,20]. In the FNA smears of an HC located in the submandibular gland, Daneshbod and Khademi found scolices, hooklets, and a portion of the laminated membrane[21]. Gupta and colleagues reported two cases in which a laminated membrane and scolices with distinctive hooklets, accompanied by an inflammatory cell background and a giant cell reaction, were found in the FNA smear of one of them. The second case had a laminated membrane surrounded by an inflammatory background[18]. The present case resembled the second case of Gupta and colleagues, wherein the aspirate exhibited cellular eosinophilic material containing a laminated membrane accompanied by an inflammatory cell infiltrate. In spite of these findings, it has been reported that the diagnostic challenge of HCs in FNA smears might arise when the features are infrequent or when inflammation is present, leading to the possibility of missed diagnoses[20]. Several scholars have discouraged the use of FNAC for HC diagnosis, particularly in endemic areas, due to the potential risks of spillage and anaphylactic reactions[22].

Regardless of the location, the definitive treatment for HC is the complete surgical excision of the cyst without any spillage. The surgical objectives are removing all parasitic elements, preventing the release of cyst contents, and preserving the affected organs as much as possible. To prevent accidental implantation, the cyst bed can be irrigated with a 3% saline solution[8,23]. The current case underwent a thoracotomy, and the cyst was totally excised.

Conclusion

Hydatid cysts can be challenging to diagnose based on imaging findings and may be misidentified as abscesses or masses. Despite the risk of anaphylaxis, FNAC may be a reliable diagnostic method if performed meticulously by an experienced radiologist.

Ethical approval

None.

Consent

Written informed consent was obtained from the patient for the 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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None.

Author contribution

R.J.R. was a major contributor to the conception of the study, as well as in the literature search for related studies. F.H.F., J.I.H. and B.A.A. conducted the literature review. H.O.A. and F.H.K. were involved in the manuscript drafting. B.A.A. and S.H.K. were involved in the critical review, and improvement of the manuscript. A.M.A. is the pathologist who performed the specimen and preparation the figure. S.H.T. is the radiologist who performed assessment of the case and preparation the figure. All of the authors were involved in the final approval of the study.

Conflicts of interest disclosure

The authors declare no conflicts of interest.

Research registration unique identifying number (UIN)

None.

Guarantor

Fahmi H. Kakamad.

Data availability statement

All data regarding this case report were included in the manuscript.

Provenance and peer review

Not commissioned, externally peer-reviewed

Patient perspective

The patient was completely satisfied with the management approach and did not have any complaints about it. Additionally, the patient praised the staff for their efforts during his treatment.

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