Primary chest wall Hydatid cyst: Review of literature with report of a new case

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1. Introduction

Hydatid cyst (HC) is a serious, endemic, parasitic disease caused by Echinococcus cestode [1]. It is endemic in sheep-farming areas of Greece, Middle East, Australasia, Sub-Saharan Africa, parts of America, and India [2]. Dog is the primary host while intermediate hosts are horse, cattle and sheep. Although humans do not share the life cycle, they may be affected accidentally. Infection is occurring either by direct contact with dog or contaminated food and fluid by eggs [2]. The most commonly affected organ is liver (52%–77%), followed by lungs (10%–40%) [3]. Rarely, it affects diaphragm, vertebrae, abdominal wall and skeletal muscles [4–6]. Hydatid disease of the chest wall is extremely rare. It might form due to either rupture of lung HC into the pleural space [3]. Primary chest wall HC is even rarer with only eight reported cases in literature [1–3,7–11].

Although it is well known that HC may affect any area of the body, reporting of cases occurring in rare organs is necessary because both presentation and clinical course of the disease differ. The aim of this study is to report a case of chest wall HC occurring in a 20-year-old pregnant lady in line with SCARE guideline with literature review [12].

1.1. Patient information

A 20-year-old, housewife, one missed period pregnant lady presented with left hypochondrial and lower chest wall painful swelling. She had this swelling for 5 years but of smaller size and no pain. In last few days, sudden increase in size occurred and became painful. They live in rural area with history of contact with domestic animals including dogs and sheeps. Her parents have been operated on for liver HC. Drug history was negative.

1.2. Clinical findings

There was 10 × 15 centimeters, tender mass with induration and overlying skin erythema, fixed to underlying chest wall.

1.3. Diagnostic assessment

The patient refused every sort of radiological examination including ultrasound because of her pregnancy. Complete blood counts and virology tests were normal.
1.4. Therapeutic intervention

Under general anesthesia, in supine position, the area was isolated by iodized pack. Oblique incision was done, on opening, clear fluid came out, with deep incision pus-like fluid and many daughter cysts drained (Fig. 1). Complete evacuation of the cyst was done with closure of the residual cavity. Drain was put. The wound closed. The procedure was performed by the second author, supervised by first author and assisted by the last author.

1.5. Follow-up and outcomes

She was advised to terminate her pregnancy and receive Anti-helminthic medication but she refused. The patient was sent home after overnight admission with oral antibiotic and analgesic. Three months later, the scar was clean and healthy, the baby was viable. Later she was los from follow up.

2. Discussion

HC which is caused by Echinococcus granulosus, transmits to humans either by direct contact with dogs or by eating contaminated food [2,3,7]. The current case reported contact with domestic animals. There is no age limit for HC affection. Admir et al. reported chest wall HC in a 9 year old male presenting with mass and chest pain for one month duration, for which benign lesion was the provisional diagnosis, while Avcı et al. reported a 72-year-old female presenting with pain [1,3]. Chest wall HC is a slow growing mass and it may be present for a long time before developing complications [9]. This lady presented with chest wall swelling for 5 years, developing hypochondrial and lower chest pain just few days before operation. According to the literatures, there are two mechanisms of primary chest wall HC. The first one is systemic spread of the embryo from the duodenal wall into the portal system or into the perigastric lymphatics which subsequently drain to the right lymphatic and the thoracic ducts and the second one local extension from the surrounding thoracic organ with disappearance of the primary source [2]. Expectoration and haemoptysis might be seen if there is involvement of pulmonary parenchyma but the current case had not have respiratory symptoms [2]. The diagnosis of HC is radiological. Indirect hemaglutination test, complimentary fixation test and dermal test may aid in diagnosis but they are neither sensitive nor specific [1,2,4,8]. Diagnosis of chest wall HC is challenging [8]. Argaw et al. performed fine needle aspiration for infraclavicular chest wall cystic lesion which showed features of lymphangioma, later exploration confirmed diagnosis of chest wall HC [2]. Alloubi and his associates managed a case of chest wall HC with provisional diagnosis of bronchogenic carcinoma invading chest wall with destruction of ribs [8]. The current case denied any sort of radiological examinations because of the pregnancy and other hematological tests were not performed as the provisional diagnosis was chest wall abscess and HC was not put in differential diagnosis. The latter was the motive for reporting this case and it is the main important learning point. Among cases of extrapulmonary thoracic HC, 2.3% reported rib destruction and full thickness chest wall involvement [9,11]. In this case, the mass was subcutaneous, partially involving muscular layers while the ribs were free from the disease. Accr et al. suggested that combined surgery and chemotherapy (Albendazole) for pre and post-operative prophylaxis provides a good outcome [2]. This case did not receive pre-operative anthelmintics and post-operative chemoprophylaxis was refused because of conception.

Table 1 shows brief literature review regarding the reported cases of chest wall HC.

| Author/ references | Age(years)/Sex | Presentation | Duration | Provisional diagnosis |
|--------------------|---------------|--------------|----------|----------------------|
| 1/Demir et al      | 9/male        | Chest pain   | 1 month  | Benign lesion         |
| 2/Argaw et al      | 36/female     | Pain, swelling | 2 years  | Lymphangiomma         |
| 3/Avcı et al       | 72/female     | Chest pain   | Not mentioned | Posttraumatic haematoma |
| 7/Tulay et al      | 48/male       | Swelling     | 5 months | Not mentioned         |
| 8/Alloubi et al    | 57/male       | Chest pain   | 3 months | Malignant tumour      |
| 9/Karapolat et al  | 69/female     | Abdominal pain | 4 years  | Hydatid cyst         |
| 10/Sarkar et al    | 58/female     | Lump         | 3 years  | Malignant tumour      |
| 11/Goyal et al     | 30/female     | Pain, swelling | Not mentioned | Not mentioned    |
In conclusion, primary chest wall HC is a very rare disease in endemic areas. Mass and pain are the most common presentation. Excision under general anesthesia is main modality of treatment.

**Patient perspective**

The patient was satisfied with the outcome of the operation.

**Conflicts of interest**

There is no conflict to be declared.

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**Ethical approval**

Approval has been taken from Kscien organization.

**Consent**

The patient gave consent for publication of the report.

**Author contribution**

Abdulwahid M. Salih, Dilshad M. Ahmed and Hanna M. Ali2: supervised and performed the operation and follow up.

Fahmi H. Kakamad, Hunar A.H and Rawand A. Essa: writing the manuscript and follow up.

**Guarantor**

Fahmi Hussein kakamad.

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