Cutaneous fistulization of the hydatid disease

A PRISMA-compliant systematic review

Zeynep Sener Bahce, MDa, Sami Akbulut, MDb,*, Ulas Aday, MDa, Fırat Demircan, MDa, Ayhan Senol, MDc

Abstract

Aim: To provide an overview of the medical literature on cutaneous fistulization in patients with hydatid disease (HD).

Methods: According to PRISMA guidelines a literature search was made in PubMed, Medline, Google Scholar, and Google databases were searched using keywords to identify articles related to cutaneous fistulization of the HD. Keywords used were hydatid disease, hydatid cyst, cutaneous fistulization, cysto-cutaneous fistulization, external rupture, and external fistulization. The literature search included case reports, review articles, original articles, and meeting presentations published until July 2016 without restrictions on language, journal, or country. Articles and abstracts containing adequate information, such as age, sex, cyst size, cyst location, clinical presentation, fistula opening location, and management, were included in the study, whereas articles with insufficient clinical and demographic data were excluded. We also present a new case of cysto-cutaneous fistulization of a liver hydatid cyst.

Results: The literature review included 38 articles (32 full text, 2 abstracts, and 4 unavailable) on cutaneous fistulization in patients with HD. Among the 38 articles included in the study, 22 were written in English, 13 in French, 1 in German, 1 in Italian, and 1 in Spanish. Forty patients (21 males and 19 females; mean age ± standard deviation, 54.0 ± 21.5 years; range, 7–93 years) were involved in the study. Twenty-four patients had cysto-cutaneous fistulization (Echinococcus granulosus); 10 had cutaneous fistulization (E. multilocularis); 3 had cysto-cutaneous-broncho-biliary fistulization; 2 had cysto-cutaneous-bronchial fistulization; and 1 had cutaneous-bronchial fistulization (E. multilocularis). Twenty-nine patients were diagnosed with E. granulosis and 11 had E. multilocularis detected by clinical, radiological, and/or histopathological examinations.

Conclusion: Cutaneous fistulization is a rare complication of HD. Complicated HD should be considered in the differential diagnosis of cases presenting with cutaneous fistulization, particularly in regions where HD is endemic.

Abbreviations: CT = computed tomography, Endo US = endosonography, ERCP = endoscopic retrograde cholangiopancreatography, HC = hydatid cyst, HD = hydatid disease, IHA = indirect hemagglutination, MRI = magnetic resonance imaging, PAIR = puncture-aspiration-injection-reaspiration, PTC = percutaneous tranhepatic cholangiography, US = ultrasonography.

Keywords: alveolar echinococcosis, complications, cystic echinococcosis, cysto-cutaneous fistulization, hydatid disease

1. Introduction

Hydatid disease (HD) is a zoonotic disease caused by Echinococcus sp. parasites (Family Taeniidae; Class Cestoda). Although 4 different Echinococcus sp. cause HD in humans, the most common are E. granulosus (causing cystic echinococcosis) and E. multilocularis (causing alveolar echinococcosis).11–16 Cystic echinococcosis, also known as a hydatid cyst (HC), is responsible for 95% of all echinococcal diseases in humans. HCs can be located in almost every organ or tissue of the body, although the liver and lungs are the most commonly involved organs. Nevertheless, the majority of patients are asymptomatic and are incidentally diagnosed by radiology performed for other reasons. Complications, such as rupture, infection, anaphylaxis, and compression of adjacent organs, may cause various clinical symptoms.21 Among the most severe complications, rupture (perforation or fistulization) is the most important. Liver HCs can rupture into the bile duct, the gastrointestinal tract, bronchi, peritoneal cavity, or pleural cavity. The progression of liver HCs into subcutaneous tissue and fistulization in the skin is a rare complication.6–12 To date, few case reports on this complication have been published. In this article, we present a case of liver HC that spontaneously fistulized into an incision on the anterior abdominal wall. We also review the relevant literature.

2. Materials and methods

According to Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines,13 a literature search was made in PubMed, Medline, Google Scholar, and Google databases using the keywords hydatid disease, hydatid cyst, cutaneous fistulization, cutaneous involvement, cysto-cutaneous fistulization, external rupture, and external fistulization alone.
or in different combinations (Flow Diagram). The language, journal, and country were not limited for this literature review. All case reports, letters to the editor, review articles, original articles, and other documents were reviewed. Reference lists of the reviewed papers were also reviewed to include citations that met the inclusion criteria. Articles without an accessible full text version, an abstract providing sufficient information, or sufficient data to compare with other studies were excluded. The following information was collected: publication year, country, publication language, paper type (full text, abstract, or unavailable), age and sex of patients, clinical presentation, Echinococcus sp. (E. granulosus or E. alveolaris), location of the fistula opening, cyst location, maximum size of cyst/lesion (mm), previous surgery, radiologic tools (ultrasonography [US], computed tomography [CT], magnetic resonance imaging [MRI], and fistulography), neoadjuvant antiparasitic chemotherapy, surgical management, postoperative antiparasitic chemotherapy, recurrence, and follow-up (months). The patient's age was given as mean ± standard deviation (SD) and range. The secondary aim of this study was to document the clinical story and management algorithm for a 56-year-old male patient who was admitted to our clinic with cysto-cutaneous fistulization caused by complicated liver HC. Local ethics committee approval was not needed because this was a retrospective literature review.

3. Results

3.1. Literature review

The literature search retrieved 38 articles involving 40 patients with cutaneous fistulization/HD involvement.[1–12,14–39] Of these, 6 articles were from India, 5 from France, 5 from Tunisia, 4 from Turkey, 4 from Morocco, 3 from Spain, 2 from Germany, 2 from Italy, and 1 each from Japan, Austria, Bulgaria, Greece, Lithuania, Romania, and Switzerland. Twenty-two articles were written in English, 13 in French, 1 in German, 1 in Italian, and 1 in Spanish. Full text was obtained for 32 of the 38 articles, whereas abstracts were available for 2 articles, and no text of any kind was available for 4 articles. The study details of the unavailable articles were obtained from 4 different articles.[5,6,14,15] Therefore, the current analysis included 40 patients (21 [52.5%] males and 19 [47.5%] females; ages, 7–93 years; mean ± SD, 54.0 ± 21.5 years). The age range of males was 29 to 87 years (mean ± SD, 57.5 ± 16.9 years) and that of females was 7 to 93 years (mean ± SD, 50.1 ± 25.0 years). The male:female ratio was 1:1. The external opening area of the fistula was reported for all patients, and the fistula opening was localized to the right hypochondrium in 11, the right flank in 4, the right thoracic wall in 4, the left hypochondrium in 3, the sternal area in 2, the inguinal region in 2, the periumbilical region in 2, the left flank in 2, and the left thoracic wall in 1 patient. Twenty-four (60%) patients had cysto-cutaneous fistulization (E. granulosus); 10 (25%) had cutaneous fistulization (E. multilocularis); 3 had cysto-cutaneous-broncho-biliary fistulization, 2 had cysto-cutaneous-bronchial fistulization; and 1 had cutaneous-bronchial fistulization (E. multilocularis). While 29 (72.5%) patients were diagnosed with E. granulosus, 11 (27.5%) patients had E. multilocularis which was detected by clinical, radiological, and/or histopathological examinations. The detailed clinical and demographic characteristics of the 40 patients with cutaneous fistulization/involvement of HD are summarized in Table 1.

3.2. Case report

A 56-year-old male patient was referred to our clinic for nausea, vomiting, and a swelling on the anterior abdominal wall. He had undergone surgery for a perforated peptic ulcer 15 years ago. A physical examination revealed hyperemia around the abdominal wall incision and a fascial defect at the abdominal midline. Abdominal US showed that a few of the lesions were consistent with HCs in both lobes of the liver and the spleen. The patient was examined with a panendoscope to differentially diagnose the dyspeptic complaints. The endoscopic examination revealed external compression of the gastric wall. The echinococcal indirect hemagglutination test was positive at a titer of 1:256. A contrast-enhanced abdominal CT scan showed 2 calcifying lesions consistent with HCs, the largest of which had a diameter of 5 cm, in the anterior and posterior segments of the right lobe of the liver. The other HC lesion was 11 × 10 cm and almost completely filled the left lobe of the liver. This HC was compressing the stomach, and was close to the anterior abdominal wall. Additionally, 2 more lesions resembling HCs, of which the larger had a 10-cm diameter, were detected in the spleen (Figs. 1 and 2). Surgery was scheduled, so the patient was administered two 400-mg doses of albendazole p.o. and a pneumococcal vaccine. Five days after the initial examination the patient presented to our emergency department with marked redness and discharge from the abdominal wall. A physical examination revealed a clear fluid discharge from a 1.5-cm opening. After local anesthesia, the orifice was dilated and a drain was inserted and a large volume of HC fluid was drained (Fig. 3). The patient underwent surgery the next day. The cyst that almost completely filled the left lobe of the liver and fistulized to the epigastrium was excised completely. Then, the cyst in the right lobe was drained with a partial cystectomy + omentectomy, and a splenectomy was performed. The fascia was closed, the incision was debrided, and the wound was allowed to heal. Albendazole was started 3 days after surgery; the patient received 4 cycles. No recurrences were detected at the 6-month follow-up visit.

4. Discussion

HD is a serious public health problem that is prevalent in many regions around the world because this is a retrospective literature review. Turkey is geographically part of the Middle East and Mediterranean regions and is one of several countries where HC disease is endemic. Humans have no role in the biological cycle of the causative organism and are accidentally infected after ingesting Echinococcus eggs in dog feces. Larvae are released into the gastrointestinal tract after the ingested eggs rupture, penetrate the intestinal wall, and pass into the portal system to reside in the hepatic sinusoids. Small-sized larvae pass through the hepatic filtration system and reach the lungs. There, the majority of larvae are cleared by a second capillary filtration system. Those that go unfiltered can easily reach many organs and systems of the body. Considering this dissemination route, it is quite easy to understand why it primarily involves the liver (50–77%) and lungs (10–40%).[1,6]

Liver HD can remain asymptomatic for years. Most asymptomatic patients are incidentally diagnosed by radiological tests performed for other indications.[6] Sometimes, HD can increase the size of the liver and patients are diagnosed by a physical examination in which a palpable liver is detected. Clinical symptoms can vary greatly depending on the size, number, and localization of HCs, their anatomical relationship with adjacent organs, and compression of adjacent organs.[16]

The most common complications of HD include secondary infections (super-infected HCs), anaphylactic reactions, vascular...
Table 1
Demographic and clinical characteristics of 38 articles involving 40 patients published in medical literature between 1987 and 2016.

| References          | Year | Country   | Language | Paper      | Age | Sex | Clinical presentation/ definitive diagnosis | Agent       | Fistula opening/ lesion location | Cyst locations | Cyst size (mm) | Previous surgery           |
|---------------------|------|-----------|----------|------------|-----|-----|--------------------------------------------|-------------|---------------------------------|----------------|----------------|---------------------------|
| Virgilio et al.     | 2015 | Italy     | English  | Full text  | 55  | F   | Cysto-cutaneous-bronchial fistula          | E granulosus| Right hypochondrion             | Right lobe-liver | 100            | Hydatid surgery            |
| Aley et al.         | 2015 | Turkey    | English  | Full text  | 80  | M   | Cysto-cutaneous fistula                    | E granulosus| Right hypochondrion             | Right lobe-liver | 60             | Liver abscess?            |
| Dalidou et al.      | 2016 | Tunisia   | French   | Full text  | 23  | F   | Cysto-cutaneous fistula                    | E granulosus| Right flank                       | NA             | 50             | NA                        |
| Mandokar et al.     | 2014 | India     | English  | Full text  | 25  | F   | Cysto-cutaneous fistula                    | E granulosus| Left hypochondrion              | Left lobe-liver | 110            | No                        |
| Jiudecius et al.    | 2014 | Lithuania | English  | Full text  | 29  | M   | Cutaneous fistula                          | E multilocular| Right flank                     | Right lobe-liver | 3              | Abscess drainage         |
| Sachdeva et al.     | 2014 | India     | English  | Full text  | 55  | M   | Cysto-cutaneous-biliary fistula            | E granulosus| Supra umbilical                 | Right lobe-liver | 50             | NA                        |
| Jayant et al.       | 2014 | India     | English  | Full text  | 65  | M   | Cysto-cutaneous fistula                    | E granulosus| Right throracic wall              | Right lobe-liver | 50             | No                        |
| Kjosev et al.       | 2014 | Bulgaria  | English  | Full text  | 60  | F   | Cysto-cutaneous fistula                    | E granulosus| Left hypochondrion              | Left lobe-liver  | 50             | Rib osteomyelitis         |
| Gupta et al.        | 2013 | India     | English  | Full text  | 50  | M   | Cysto-cutaneous fistula                    | E granulosus| Left flank                       | Liver- biliary   | 180            | No                        |
| Bouassida et al.    | 2012 | Tunisia   | French   | Full text  | 24  | F   | Cysto-cutaneous fistula                    | E granulosus| Right hypochondrion              | Intrapitoneal   | 80             | Hydatid surgery           |
| Hamid et al.        | 2012 | India     | English  | Full text  | 7   | F   | Cysto-cutaneous fistula                    | E granulosus| 8th intercostal space              | Right lobe-liver | 50             | Hydatid surgery           |
| Lacetona et al.     | 2012 | Italy     | Italian  | Full text  | 60  | F   | Cysto-cutaneous fistula                    | E granulosus| Left flank                       | Kidney-left     | 105            | No                        |
| De Lewaissiere et al.| 2012| France   | French   | Full text  | 40  | F   | Cysto-cutaneous fistula                    | E granulosus| Infernal, left                    | Pelvis          | 70             | No                        |
| Ben Amour et al.    | 2014 | Tunisia   | French   | Full text  | 33  | M   | Cysto-cutaneous fistula                    | E granulosus| Right flank                       | Right lobe-liver | 50             | No                        |
| Prieto-Neto et al.  | 2011 | Spain     | English  | Full text  | 58  | F   | Cysto-Cutaneous-Biliary fistula            | E granulosus| Right hypochondrion              | Right lobe-liver | 50             | Hydatid surgery           |
| Ramia Angeli et al. | 2011 | Spain     | Spanish  | Full text  | 76  | M   | Cysto-cutaneous fistula                    | E granulosus| Left hypochondrion               | Intrapitoneal   | 80             | Hydatid surgery           |
| Koner et al.        | 2011 | India     | English  | Full text  | 68  | M   | Cysto-cutaneous fistula                    | E granulosus| Umbilicus                        | Right lobe-liver | 120            | NA                        |
| Martin-Perez et al. | 2011 | Spain     | English  | Full text  | 87  | M   | Cysto-cutaneous fistula                    | E granulosus| Right hypochondrion              | Right lobe-liver | 180            | No                        |
| Schmoldt et al.     | 2010 | Germany   | English  | Full text  | 71  | F   | cutaneous fistula                          | E multilocular| Epigastrium                      | Left lobe-liver  | 75             | No                        |
| Yakan et al.        | 2009 | Turkey    | English  | Full text  | 93  | F   | Cysto-cutaneous fistula                    | E multilocular| Infra umbilical                 | Left lobe-liver  | 90             | Cholecystectomy          |
| Ali et al.          | 2009 | Morocco   | French   | Full text  | 45  | M   | Cysto-cutaneous fistula                    | E granulosus| Right hypochondrion              | Right lobe-liver | 50             | NA                        |
| Chaifki et al.      | 2009 | Morocco   | French   | Full text  | 35  | M   | Cysto-cutaneous fistula                    | E granulosus| Left thoracic wall                | Left lobe-liver  | 60             | No                        |
| Onat et al.         | 2008 | Turkey    | English  | Full text  | 21  | F   | Cysto-cutaneous fistula                    | E granulosus| Right thoracic wall               | Right lobe-liver-pulmonary | 40            | Hydatid surgery           |
| Florea et al.       | 2008 | Romania   | English  | Full text  | 71  | F   | Cysto-cutaneous fistula                    | E granulosus| Right hypochondrion              | Right lobe-liver | 60             | No                        |
| El Ammari et al.    | 2008 | Morocco   | French   | Full text  | 64  | M   | Cysto-cutaneous fistula                    | E granulosus| Infranal, right                   | Kidney-right    | 340            | No                        |
| Salorafas et al.    | 2006 | Greece    | English  | Full text  | 85  | M   | Cysto-cutaneous fistula                    | E granulosus| Right hypochondrion              | Right lobe-liver | 50             | NA                        |
| Kismet et al.       | 2006 | Turkey    | English  | Full text  | 43  | M   | Cysto-cutaneous fistula                    | E granulosus| Epigastrium                      | Spleen          | 95             | Cholecystectomy          |
| Bastel et al.       | 2005 | France    | French   | Full text  | 42  | M   | Cysto-cutaneous fistula                    | E granulosus| Epigastrium                      | Liver           | 140            | Hydatid surgery           |
| Grisy-Guilla et al. | 2004 | France    | French   | Full text  | 39  | F   | Cysto-cutaneous fistula                    | E granulosus| Epigastrium                      | Right lobe-liver | 40             | No                        |
| Reuter et al.       | 2003 | Germany   | English  | Full text  | 60  | M   | cutaneous fistula                          | E multilocular| Stomal area                       | Liver, thoracic wall | 40            | Hydatid surgery           |
| Selmi et al.        | 2001 | Tunisia   | French   | NA         | 11  | F   | Cysto-cutaneous-bronchial fistula          | E granulosus| Right thoracic wall               | Right lobe-liver | NA             | NA                        |
| Ambo et al.         | 1999 | Japan     | English  | Full text  | 68  | M   | Cysto-cutaneous fistula                    | E multilocular| Right hypochondrion              | Liver           | NA             | Hydatid surgery           |
| Harandou et al.     | 1997 | Morocco   | French   | Abstract   | 70  | F   | Cysto-cutaneous-bronchial fistula          | E granulosus| Right flank                       | NA             | NA             | NA                        |
| Bresson-Hadini et al.| 1996| Austria   | English  | Full text  | 68  | M   | Cysto-cutaneous fistula                    | E multilocular| Epigastrium                      | Liver           | NA             | No                        |
| Berthet et al.      | 1992 | France    | French   | NA         | 41  | M   | Cutaneous fistula                          | E multilocular| Right hypochondrion              | Right lobe-liver | 50             | NA                        |
| Golematitis et al.  | 1991 | France    | French   | NA         | 71  | F   | Cysto-cutaneous fistula                    | E multilocular| Right hypochondrion              | Right lobe-liver | 50             | NA                        |
| Tschudi et al.      | 1988 | Switzerland| German  | Abstract   | 47  | M   | Cutaneous fistula                          | E multilocular| Right thoracic wall              | Liver           | NA             | NA                        |
| Kehila et al.       | 1987 | Tunisia   | French   | NA         | 86  | F   | Cutaneous-bronchial fistula                | E multilocular| Right flank                       | Right lobe-liver | NA             | NA                        |
| References  | Radiologic tools | Neoadjuvant treatment | Surgical management | Adjuvant treatment | Recurrence | Follow-up (mo) |
|-------------|------------------|-----------------------|---------------------|-------------------|------------|----------------|
| Virgillio   | CT + Fistulography | No                    | Nonoperative management | Yes-3 mo         | No         | NA             |
| Aley        | US + CT           | NA                    | Drainage            | NA                | No         | 6              |
| Dabdou      | US + CT           | No                    | Excision of fistulous tract + pericystectomy | No              | No         | 132            |
| Mandelkjar  | US + X-Ray        | No                    | Excision of fistulous tract + pericystectomy | ABZ-2 mo        | No         | 4              |
| Jooolekis   | US + CT           | ABZ                   | Excision of fistulous tract + pericystectomy (6 + 7) | ABZ-12 mo       | No         | 12             |
| Sachdeva    | CT + Fistulography | NA                    | Thoracotomy + excision of cyst cavity + diaphragmatic repair + tube drainage | NA             | NA         | NA             |
| Jayant      | US + CT           | ABZ                   | Right hepatectomy + excision of fistulous tract | ABZ             | No         | 12             |
| Koseev      | CT                | NA                    | Percystectomy + excision of fistulous tract | ABZ             | No         | 12             |
| Gupta       | US + CT           | NA                    | Excision of cyst cavity + thoracotomy + diaphragmatic plication | NA             | No         | Died           |
| Bouassida   | CT                | NA                    | Total cystectomy + excision of fistulous tract | ABZ-6 mo        | No         | 60             |
| Hamid       | US + CT + Fistulography | NA            | Excision of fistulous tract + evacuation of cyst cavity | ABZ-6 mo        | No         | 5              |
| Laxotera    | CT                | NA                    | Excision of fistulous tract + left nephrectomy | ABZ             | No         | 24             |
| De Leusseire| US + CT           | ABZ                   | Nonoperative management | ABZ             | No         | 5              |
| Ben Ameur   | US + CT           | ABZ                   | Excision of fistulous tract + partial cystectomy | ABZ-6 mo        | No         | 18             |
| Pinto-Merio | CT + Fistulography | NA                    | Tissucol sealing injection in fistulous tractus | No             | No         | 24             |
| Ramia Angel | CT                | NA                    | Excision of fistulous tract + total cystectomy (peritoneal and subcutaneous) | ABZ-3 mo        | No         | NA             |
| Korner      | US                | ABZ                   | Partial cystectomy + excision of fistulous tract + total cystectomy (peritoneum) | ABZ-2 mo        | No         | 60             |
| Martin-Perez| US + CT           | ABZ                   | Excision of fistulous tract + total cystectomy (subcutaneous cyst) + partial cystectomy (liver) | NA             | No         | NS             |
| Schmoldt    | CT                | MBZ + ABZ             | Excision of fistulous tract + left hepatectomy + partial pericardial + diaphragmatic Resection | ABZ             | No         | 12             |
| Yaken       | CT                | ABZ                   | Nonoperative management | ABZ             | NA         | NA             |
| A1          | CT                | NA                    | Excision of fistulous tract + partial cystectomy | ABZ-3 mo        | No         | 18             |
| Chatik      | CT                | NA                    | Resection of the lateral arch of the 6th rib | ABZ-6 mo        | No         | 18             |
| Onat        | CT                | NA                    | Cystotomy + cephalonage | ABZ             | No         | 3              |
| Florea      | US + CT           | No                    | Excision of fistulous tract + partial cystectomy | ABZ + Praziquantel- 6 mo | No | 36             |
| El Ammari   | CT                | NA                    | Percystectomy | NA             | NA         | NA             |
| Sakorafas   | CT                | NA                    | Partial cystectomy + subcutaneous cyst excision | NA             | No         | 12             |
| Kismet      | US                | NA                    | Partial cystectomy | ABZ             | No         | 3              |
| Barleto     | US + CT           | NA                    | Percutaneous drainage | ABZ             | Regressed | 3              |
| Grig-Guilla | US + CT + MR      | ABZ                   | Excision of fistulous tract + partial cystectomy | ABZ-1 mo        | No         | NA             |
| Rauter      | CT + MRI + FDG PET-CT | Amphotericin B     | Nonoperative management | NA             | Successfully | 14             |
| Rauter      | CT + MRI + FDG PET-CT | Amphotericin B     | Nonoperative management | NA             | Decreased | 25             |
| Selmi       | US                | NA                    | Cystectomy | NA             | No         | 9              |
| Ambo        | CT                | NA                    | Excision | NA             | No         | 36             |
| Hassanou    | NA                | NA                    | Excision of fistulous tract + thoracotomy + pericystectomy | NA             | NA         | NA             |
| Bresson-Hadni| X-Ray             | ABZ                   | Nonoperative management | ABZ             | No         | 2              |
| Bresson-Hadni| CT + Fistulography | ABZ                   | Nonoperative management | ABZ             | Regressed | 12             |
| Berthet     | US                | NA                    | Partial cystectomy | NA             | No         | 84             |
| Golematis   | CT + Fistulography | NA                    | Partial cystectomy + drainage | NA             | No         | 24             |
| Tschedu     | CT                | NA                    | Wide local excision | NA             | No         | 48             |
| Kehila      | US + Fistulography | NA                    | External catheter drainage | Antibiotherapy | NA         | NA             |

**Note:**
- **ABZ=** albendazole, **CT=** computed tomography, **E granulosus=** Echinococcus granulosus, **E multilocularis=** Echinococcus multilocularis, **F=** female, **M=** male, **MBZ=** mebendazole, **MRI=** magnetic resonance imaging, **NA=** non-available, **US=** ultrasonography.
Figure 1. Axial abdominal computed tomography images obtained from different sections after injecting contrast material. A lesion consistent with a stage III hydatid cyst is seen in the posterior segment of the right liver lobe and another lesion consistent with a stage V hydatid cyst is seen in the anterior segment of the right liver lobe. Lesions consistent with stage I and stage V hydatid cysts are seen in the upper and lower poles of the spleen, respectively.

Figure 2. Sequential abdominal computed tomography images taken after injecting contrast material. A lesion consistent with a hydatid cyst is seen starting from segment 3 of the left liver lobe and protruding into the anterior abdominal wall.
Clinicians specify the extension of the fistula tract and bronchobiliary structures by fistulography in a patient who had undergone IHC surgery years ago. Prieto-Nieto et al reported that they filled the fistula tract with Tissucol, a biological fibrin glue, and successfully treated the patient. In conclusion, the success rate of fistulography for showing organ involvement of a cystocutaneous fistula is 85.7%. In the present case, we did not use fistulography due to discharge of bouses of daughter vesicles through the incision scar. However, fistulography can be performed under appropriate conditions in all cases with a narrow fistula orifice and an unclear diagnosis. We cannot provide a clear recommendation due to a lack of sufficient data on this subject.

The most commonly used serological tests for diagnosing complicated and uncomplicated HD and monitoring posttreatment recurrences are enzyme-linked immunosorbent assay, indirect hemagglutination, serum immunoelectrophoresis, the complement fixation test, and immunofluorescence assay. Nineteen of 40 enrolled patients underwent various serological tests; 94.7% had a positive test result. Only 1 patient returned negative serology, although costal HD was shown histopathologically.

The most appropriate strategy to minimize recurrence in patients with complicated HD and cutaneous fistulization is to combine surgical and medical treatment modalities. A 2 to 4-week neoadjuvant medical treatment followed by elective surgery is the most appropriate approach for patients who are not in need of urgent surgical intervention. Surgical treatment includes en bloc resection of the primary hydatid lesions causing the complication, diseased skin region, and fistula tract. Radical options, such as pericystectomy, segmentectomy, and lobectomy, can be used to minimize recurrence. Adjuvant medical treatment is not necessary in patients treated with radical surgery.

HD follows several stages before reaching the abdominal or thoracic wall to develop into an external rupture. Stage I hydatid lesions protrude into the innermost muscular layer of the abdominal or thoracic wall. Stage II lesions pass beyond the muscular layer and protrude into subcutaneous soft tissue. Stage III is characterized by the passage of lesions into subcutaneous tissue and their fistulization in the skin, which is called an internal rupture (gastric outlet obstruction or duodenal compression), and can be easily made in almost all patients. A histopathological examination of the fluid discharged from the external orifice of a fistula can demonstrate the parasite itself and identify a superinfection, if present. US, CT, and MRI are the most commonly utilized radiological tools to diagnose cutaneous involvement and locate lesions causing complications of the HD.

Endoscopic retrograde cholangiopancreatography (ERCP), percutaneous transhepatic cholangiography (PTC), and endosonography are the other invasive radiological methods used for diagnostic and therapeutic purposes. The most useful radiological method for cutaneous manifestations of the disease is contrast-enhanced fistulography. This technique helps clinicians specify the extent of the fistula, the location and size of a fistulized lesion, and its relationship with bile ducts, bronchopleural structures, and the pelvicalical system. In the present study, 7 (17.5%) of the enrolled patients underwent fistulography in addition to other radiological examinations. Bresson-Hadni et al reported that fistulography failed to show any connection between the fistula tract and the liver lesion. Prieto-Nieto et al showed a connection between the fistula tract and bronchobiliary structures by fistulography in a patient who had undergone IHC surgery years ago. Prieto-Nieto et al reported that they filled the fistula tract with Tissucol, a biological fibrin glue, and successfully treated the patient. In conclusion, the success rate of fistulography for showing organ involvement of a cystocutaneous fistula is 85.7%. In the present case, we did not use fistulography due to discharge of bouses of daughter vesicles through the incision scar. However, fistulography can be performed under appropriate conditions in all cases with a narrow fistula orifice and an unclear diagnosis. We cannot provide a clear recommendation due to a lack of sufficient data on this subject.
cutaneous fistulization, as a limited number of such cases have been reported so far. This topic should be explored further in future studies.

4.1. Topic highlights

- Cutaneous fistulization is a rare but serious complication of hydatid disease.
- A preoperative diagnosis is quite challenging when no cyst material has been drained from the external os of a fistula tract.
- Awareness is an important approach to hydatid disease and its complications.
- Complicated hydatid disease should be considered in the differential diagnoses of patients presenting with cutaneous fistulization, particularly in regions where hydatid disease is endemic.

References

[1] Mandalkar SD, Ramakanth B, Anil Kumar PL, et al. Cystocutaneous fistula of the left lobe of liver: an extremely rare presentation of hydatid liver cyst. Int Surg J 2015;2(2):109–11.
[2] Jayant K, Agrawal S, Agarwal R, et al. Spontaneous external fistula: the rarest presentation of hydatid cyst. BMJ Case Rep 2014;2014: pii: bcr2014203784.
[3] Juodeikis Z, Pusksis T, Senin D, et al. Echinococcus multilocularis infection of the liver presenting as abdominal wall fistula. BMJ Case Rep 2014;2014: pii: bcr2014203769.
[4] Sachdeva V, Singh SK, Sood V, et al. Cutaneous bronchobiliary fistula following laparotomy for ruptured hydatid cyst of the liver. Int Surg J 2014;1(1):155–7.
[5] Kjossev KT, Teodosiev IL. Cutaneous hydatid disease. BMJ Case Rep 2014;2014: pii: bcr2014203784.
[6] Korwar V, Subhas G, Gaddikeri P, et al. Hydatid disease presenting as the rarest presentation of hydatid cyst. Apropos of a case. J Chir (Paris) 1997;134:31–4.
[7] Harmil R, Shera AH, Bhat NA, et al. Hydatid cyst of liver: Spontaneous cutaneous cutaneous cutaneous cutaneous cutaneous fistula of liver. J Am Acad Dermatol 1996;34(5 Pt 2):873–9.
[8] Bouassida M, Sassi S, Mighri MM, et al. Parietal complications of hydatid cyst of liver. Report of two cases in Tunisia. Bull Soc Pathol Exot 2012;105:259–61.
[9] Kpitit J, Dousset B, Bouchiha A, et al. Hydatid cyst of the liver: diagnosis and management. J World Natl Cystic Fibrotic Dis 2011;96:69–73.
[10] Harandou M, el Idrissi F, Alaziz S, et al. Spontaneous cutaneous cysto-intrahepatic bile duct fistula: the rarest presentation of hydatid liver cyst. BMJ Case Rep 2014;2014: pii: bcr2014203784.
[11] Bresson-Hadni S, Humbert P, Paintaud G, et al. Skin localization of alveolar echinococcosis. Antimicrob Agents Chemother 2003;47:3586–91.
[12] Martin-Perez E, Gomez J, Rubio I, et al. Spontaneous cutaneous cysto-cutaneous fistula of liver echinococcal cyst. Ann Trop Med Parasiato 2006;100:51–5.
[13] Prieto-Nieto MI, Perez-Robledo JP, Alvarez-Luque A, et al. Cutaneous fistulization associated with hepatic cystic echinococcosis. Springerplus 2015;4:538.
[14] Prieto-Nieto MI, Perez-Robledo JP, Alvarez-Luque A, et al. Cutaneous bronchobiliary fistula treated with Tissucol sealant. Cardiovasc Intervent Radiol 2011;34(suppl 2):S32–5.
[15] Cano A, Marim E, Rabago J, et al. Hydatid cyst of the kidney fistulized at the skin-case report. J Maroc Urol 2008;9:38–41.