Unusual presentation of hydatid disease
5 cases collected in a period of 4 years in 2nd March Hospital Sebha

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Summary:
A 4 years prospective collection of 5 cases who were presented in surgical department with symptoms & signs not suggesting hydatid disease which were diagnosed intra-operatively supported by histopathological examination.

Key words:
Hydatid disease, unusual presentation, histopathology.

Introduction:
Echinococcus granulosus is a protozoan of a head and 3 segments, its end host is the bowel of canine animals who got the cadaver or remains of herbivorous animals which not eliminated properly when a cysts founds in it, usually its liver.

Bets, eating of improperly cleaned raw vegetables are the common methods of infestation with the parasite, its intermediate stage form a cyst (hydatid) which derived from Greek and means watery vesicle .

Internationally, hydatid disease is endemic in Mediterranean sea bordering countries, south America and Oceania, nationally the disease is endemic in north west of Libya ,and Aljabal Alakhder zones.1

We though, because it’s a Sahara –no sheep rearing area– Fazzan region is less likely to be involved, but many cases of hydatid disease were detected, commonly of liver and lungs (the two common site of infestation) but during the period of 2000–2004 cases with presentations not suggesting hydatid disease were encountered.

Reviewing the literatures of rare and unusual cases of hydatid disease were searched for.

Material and methods:
During the 4 years period (may 2000 to October 2004) 5 patients admitted to surgical department of 2nd March General Hospital, three patients presented with acute abdomen of varying severity, with vague clinical findings not helping in reaching a definitive diagnosis, the 4th patient presented with neck swelling and dysphagia, the 5th patient was presented with right upper thigh swelling.

case 1:
67 years old lady suffering from difficulty of swallowing for few months ago which become progressive, on admission the patient was only on liquid diet.

Fig. (1)
The swelling at floor of the mouth extending outward was present for 2 years ago and became more prominent recently.

Fig. (2)

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A part of leucocytosis (eosinophilia) the rest of investigations were normal ultrasound examination reveal a cystic lesion with well defined membrane CT scan reveal a hypodense lesion with clear inside, no calcification in the lesion. Fig (1, 2).

During operation, a clear fluid aspirated, the dissection of the cyst off the floor of the mouth, a whitish membrane was extracted from the cystic cavity. Histopathology report revealed Echinococcus membrane.

The patient had coarse of albendazole, and 1 year follow up prove no hydatid disease any where.

Case 2:
A boy child of 12 years old was playing football with his friends when he was colloid with another player. The child presented with abdominal pain, tenderness at left side of his chest.

Clinically, the child status was stable but surprisingly he was flashed, puffy face, warm, and had attacks of scratch episodes, he was tachycardiac 110 b/m, blood pressure was 110/70, Hb value was 13.6g% with eosinophilia, chest X ray reveal normal chest.

Ultrasound of the abdomen reveal free fluid in peritoneal cavity with rupture spleen which contain a cyst.

Case 3:
A girl of 3 years old presented in pediatric department compailing of diffuse abdominal pain for 2 days, it was colicky, no vomiting nor diarrhea, exaggerated bowel movement x ray reveal multiple fluid levels indicating mechanical small bowel obstruction. Fig (3).

As a part of preparation for surgery chest X ray reveal a cystic lesion in lower zone of the right.

Fig (4) At Laparatomy; segments of lower part of jejunum is adherent to thick firm glomeronated mass on the urinary bladder and fundus of the uterus which was free posteriorly.

Fig. (3)

During dissection of the mass a whitish membrane bulged out partial aspiration was performed, the membrane removed, the bowel continuity was maintained.

Histopathology of the membrane was proved to be Echinococcus granulosus.

Fig. (4)
Case 4:
A young girl of 10 years old, present to pediatrics department complaining of central abdominal pain, vague in nature, this vague pain was on and off for the last 1 year. A part of fullness at right side of the abdomen no other clinical finding, all her investigations were within normal a part of eosinophilia, radiological studies were normal except ultrasound finding of a cystic lesion extending from lower posterior surface of the liver. At Laparotomy; the liver, spleen, and bowel were normal, soft cystic swelling pushing the liver, hepatic flexure and posterior peritoneum anteriorly and to the left side, partial mobilization of the liver and ascending colon. During mobilization of the mass it rupture and yellowish fluid aspirated followed by a whitish membrane which was withdrawn from the cavity which thoroughly irrigated, drain was kept for 4 days. The child had a good uncomplicated recovery.

Case 5:
Sheep keeper 55 years old presented with a firm painless mass at upper lateral aspect of the left thigh, which has been noticed since 3 months. All the investigations were normal, radiological study reveal normal femur, vague soft tissue mass which proved by ultrasound to be cystic in nature. Clear dissection of the mass, the cyst wall identified the cyst aspirated, opened a whitish membrane was removed, the cavity drained. Histopathology examination confirm it as Echinococcus granulosus.

Discussion:
Hydatid disease continues to be a medical and sanitary problem in endemic areas, and is being observed more frequently than expect in some area where not seen before. Classical presentation and findings are the rule, but non usual location sites should be thought for proper management. As the egg of Echinococcus swallowed it hatch and penetrate to portal circulation and will be filtered to the two common filter sites; liver or lungs, but from theoretical point of view any anatomical structure may be affected. The usual thinking of the common diseases in specific organs in an endemic area of hydatosis may pose a difficult clinical situation specially if primary hydatid disease is detected or suspected; thyroid hydatid disease may present as a cold nodule and a cystic lesion which its usual therapy is aspiration cytology and an anaphylactic reaction or dissemination may occur. Spleen involvement like our case produced a diagnostic error as a benign seplnic cyst, same problem in case of kidney hydatosis. In the literatures, other than liver and lung sites in humans, with their usual presentations and obvious on chest X-ray and ultrasound, hydatid disease involves the liver in approximately 75% of cases, the lung in 15% and other anatomic locations in 10% detected either as primary sites or with unusual presentations, which include; pectoralis major, brain, spleen, pancreas, ovaries, breast mass, adrenal gland and mediastinum, hydatid cyst in most of time create its symptoms by its pressure on the surrounding leading to organ function disturbance and obstruction. Diagnosis of echinococcosis in endemic areas and with common presentations usually a straightforward. But when an unusual symptoms in cases where a primary hydatid disease detected, a thorough investigation should be conducted in order to reach a definitive diagnosis. Many modalities of investigative tools and methods applied in order reach a definitive diagnosis.

Ultrasound examination is most common non invasive technique, easy to apply and widely used with accuracy of (80% in cystic parasitic lesion) & (66.4% in solid parasitic lesion). MRI and CT scan play a significant role in non invasive diagnosis with accuracy of 80–93% of the cases. Serology tests that is very helpful in diagnosis of echinococccosis, yet those tests are not very accurate; Casoni test; the 1st test used, was and still available test and widely used, but is not accurate and with major cross reaction with other reactions. The indirect haemagglutination test (IHT); being less sensitive but more specific than the radiological diagnosis, it was positive in 80-90%, in a Yemeni study utilizing anti-hydaitid cyst antibodies, a sensitivity of ( ) & a specificity ( ) had been recorded. An enzyme-linked immuno-electrotransfer blot (EITB), which have a higher sensitivity and specificity than IHT exceeding 90%, verified using a western-blot method in order to detect the reaction with 8 kDa fraction of Echinococcus antigen, the aid of Em2 plus or Em-18 ELISA tests which detect antibodies specific for E. multilocularis. The fine needle aspiration (an invasive method) provides accurate diagnosis nearly
100%; as it provides the intra-cystic fluid for electrolytes, scoleces and antigen antibodies immune reaction. Ultrasound as an assisting tool for defining and localizing the cyst play a major role for FAB.\textsuperscript{24,25}

**Therapy:**
Surgery remain the main stay of therapy, the traditional method of cystopericystectomy - the method of choice - in which the cyst membrane is removed after using scolidal agents, hypertonic saline is commonly used in order to kill the germinal layer to prevent dissemination during surgery.\textsuperscript{29}

Many modalities of therapy have been developed, chemotherapy, by using albendazole either alone or with mebendazole is the common drug in use for hydatidosis usually before and after surgery.\textsuperscript{30} Albendazole has been studied as a sole treatment of hydatidosis with limit value as the rate of recurrence was up to 78.5% within 2 years. Still applied as treatment in cases where surgery is contraindicated.\textsuperscript{31} Death of germinal epithelium usually followed by its detachment from the adventitial layer and decrease of cyst size; Fig (5).

Ultrasound is the method of choice in assessing these changes. Puncture, aspiration, scolidal injection, and re-aspiration (PAIR) technique has been used as a modality of treatment of hydatid cyst.\textsuperscript{32} With good results, no recurrences, the only complication encountered was abscess formation in the residual cavity, with development of minimal invasive techniques, a combination procedures developed utilizing this method with good results, rare complications and a long term follow up showed that the procedure is safe, effective.\textsuperscript{33,34}

Follow up of hydatid disease patients treated by any of mentioned methods by immunological methods prove of no value as the values remain either same or changed insignificantly.\textsuperscript{35}

Still the principle of (prevention is better than cure) applicable for hydatid disease control, good sanitary, proper & clean hygiene and animal control are and still the used methods of control of hydatidosis in Australia a vaccination of intermediate host by applying EG95 to the goats and sheep prove to be effective,\textsuperscript{36,37} could have wide applicability as a new tool for use in hydatid control campaigns.

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**Fig (5)**

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