CASE REPORT

Unusual sites for primary hydatid cysts: self experience with five cases

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SUMMARY

Hydatid disease is a zoonotic disease caused by the tapeworm *Echinococcus granulosus*. It is common in the sheep-raising countries including Iraq. The usual site for involvement is the liver, followed by the lungs. Other sites may be affected less commonly and require a high index of suspicion for diagnosis. We present our experience with five cases of unusual sites of primary hydatid cyst, including the pancreas, the abdominal wall, the spleen, the back and the thigh. Three patients were females and two patients were males; their ages were between 15 and 39 years. All the patients were operated at our centre, and after a period of follow-up ranging between 2 and 6 years, there was no reported recurrence in any of the patients, neither at the primary site nor at other sites.

BACKGROUND

Hydatid disease is caused by the tapeworm *Echinococcus granulosus*. Dogs are the definitive hosts while humans and sheep act as intermediate hosts. The dog gets infected by eating the viscera of sheep that contain hydatid cysts. The scolices which are present inside the cysts become anchored to the wall of the small intestine of the dog and grow to become adult taenia and start to produce ova into the bowel. Humans and sheep get the infection by eating vegetables and grass contaminated with ova from dogs' faeces, that is why hydatid disease is most commonly found in sheep-raising areas, including South Australia, New Zealand, Africa, Greece, Spain and the Middle East, including our country, Iraq.¹

Human hydatid cyst can occur at any site of the body. After ingestion of the ova, it penetrates the

Figure 2 Operative image showing the hydatid cyst in the pancreas.

intestinal mucosa and is carried by portal circulation to the liver, where it develops into an adult cyst. Most cysts are caught in the hepatic sinusoids; therefore, 50–70% of hydatid cysts form in the liver. The ova which pass the liver reach the lungs, especially the right lung which is the second most commonly affected organ. In fact, the liver and lung-hydatid disease represent more than 90% of the cases. Other organs are reported to be involved less commonly, the exact incidence is difficult to be ascertained; these organs include the kidneys, heart, spleen, central nervous system, eyes, muscles, breast and ovaries. Haematogenous or lymphatic spread could account for these unusual primary lesions.²

CASE PRESENTATION

Case 1: A 22-year-old woman presented to us with 6 months history of vague upper abdominal pain, which occurred mainly after meals. The pain was increasing in intensity and was associated with nausea and vomiting a few times. She started to feel a mass in the epigastrium during the past

Figure 1 CT scan demonstrating a cyst in the body of the pancreas. A picture highly suggestive of a hydatid cyst.

Figure 3 Operative image showing the cyst contents being evacuated.

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Figure 4 CT scan showing a cystic lesion related to the abdominal wall, with a septated appearance; the picture was highly suggestive of hydatid cyst.

2 months which was increasing in size. She had no history of gall stones or alcoholism. On examining the patient, she was not jaundiced; there was a tender, soft abdominal mass in the epigastric region, and other aspects of the examination were unremarkable.

All laboratory investigations were normal. Ultrasonography revealed a cystic mass behind the stomach, multiloculated 7*15 cm; CT scan (figure 1) revealed the mass to be occupying the whole body and tail of the pancreas and closely adherent to the posterior wall of the stomach, and revealed a septated pattern and internal echoes which were highly suggestive of the diagnosis of hydatid cyst. The diagnosis was proved by serology and there were no other cysts in other organs. The patient was operated through gastrotomy, with an evacuation of the contents of the cyst (figures 2 and 3) and cyst gastrotomy.

Case 2: A 27-year-old healthy woman presented with 3 months history of an enlarging painless mass at the left side of the abdomen below the costal margin; her medical, surgical and family history were unremarkable. On examining the mass, it was

Figure 5 Operative image showing the hydatid cyst in the spleen.

Figure 6 Operative image showing the hydatid cyst in the back of torso.

cystic, oval in shape, not tender and clearly extraperitoneal. CT scan (figure 4) revealed the mass which was 10*17 cm, related to the left side of the abdominal wall with internal septa—a picture which was highly suggestive for the diagnosis of hydatid cyst of the abdominal wall. The diagnosis was proved by serology and there were no other cysts in other organs. The patient was operated by complete cystectomy.

Case 3: A 27-year-old woman presented with a history of left sided, dull, aching abdominal pain for 1 month. There were no remarkable findings on examination. Her laboratory investigations were normal. Ultrasonography and CT scan demonstrated two cystic lesions in the spleen measuring 4*3 and 2*4 cm, with internal echoes. The diagnosis was proved by serology and there were no other cysts in other organs. On surgery, the patient was found to have two hydatid cysts of the spleen (figure 5). Splenectomy was done for her.

Case 4: A 15-year-old healthy boy was referred to us with a clinical diagnosis of a lipoma in the back of torso. The lump was present for the past 2 years but increased in size during the past month and made the patient seek advice for it. The mass was cystic on examination and it felt deep to the muscles. Ultrasonography raised suspicion of a cystic lesion, which on MRI of the back was suspected to be a hydatid cyst. The diagnosis was proved by serology and other parts of the body were carefully investigated for a primary lesion. The cyst was operated by complete cystectomy (figure 6).

Figure 7 The site of the mass in the upper part of the inner thigh.

Figure 8 Operative image showing the mass being deep to the muscles.

Case 5: A 39-year-old otherwise healthy man presented to us from the outpatient clinic with a painless swelling of the upper right thigh, which was gradually increasing in size with little discomfort. On examination (*figure 7*), the lump was located at the medial aspect of the right thigh; it was oval in shape, 15×15×10 cm, firm in consistency, slightly mobile, without fluctuation and not attached to the overlying skin which was normal in appearance. Ultrasonography and MRI raised a high suspicion of a hydatid cyst located in the posterior compartment of the thigh. The diagnosis was proved by serology and other parts of the body were carefully investigated for a primary lesion.

The patient was operated with an S shape incision made over the mass (*figure 8*), which was discovered to be deep to the muscles (*figure 9*). Accidental rupture occurred during dissection and revealed an infected hydatid cyst with too many daughter cysts. Removal of all daughter cysts and all the layers of the cyst was done (*figure 10*) and the wound was closed with a suction drain inserted that was removed on the third post-operative day.

INVESTIGATIONS

On suspicion of hydatid disease, all our patients were carefully investigated to exclude the presence of hydatid cyst in other

Figure 9 Operative image showing the cyst lying in between the muscles of the thigh.

Figure 10 Operative image showing excision of the cyst following accidental rupture.

parts of the body, especially the liver by ultrasonography and CT of the abdomen. Pulmonary hydatidosis was excluded in all patients by chest imaging (X-ray and CT of the chest). Brain MRI was performed in two patients who had history of headache to exclude brain hydatid disease. Serological confirmation was done in all cases using ELISA.

OUTCOME AND FOLLOW-UP

All patients had an uneventful postoperative recovery with no reported morbidity during the follow-up period ranging between 6 years in the first case and 2 years in the last one.

DISCUSSION

Hydatid disease is still a common and important problem in our country with a great impact upon the health system. The reported incidence of hydatid disease in Iraq is between 1 and 12/100 000 of the population and varies according to the area of study with highest reports in the southern parts.^{3–7}

In our centre, the workload for surgically treated hydatid disease between the years 2007 and 2013 was between 70 and 85 cases per year for each surgeon excluding neurological hydatid disease. This reflects the high prevalence of the condition and the experience of our surgeons to deal with the disease. More than 90% of the surgical cases of hydatid disease at our centre were located at the liver, lungs or both. Beside our five cases, four other cases of unusual locations were reported during the same period including primary hydatid disease of the breast, eye, common bile duct and femur.

Hydatid disease at unusual sites carries a diagnostic challenge even in endemic areas like our country. A high index of suspicion is required, especially in patients coming from rural areas who give a long history of contact with domestic animals, especially dogs and sheep.

Hydatid disease of the pancreas is extremely uncommon. The cyst is asymptomatic in most cases except when causing obstruction of pancreatic duct or common bile duct. Large cysts may cause abdominal pain. Diagnosis can be readily made by ultrasonography and abdominal CT; the picture is usually clear but sometimes the cyst can look like a cystic tumour which should be excluded.

Our patient had upper abdominal pain and was diagnosed preoperatively by the radiological and serological picture. We selected to perform a cystogastrostomy because of the location

of the cyst at the neck and body of the pancreas and the dense adhesion with the posterior wall of the stomach and a suspected connection of a pancreatic duct within the cyst. Cysts in the body and tail can be treated by resection.⁸

Hydatid cyst of the spleen is very uncommon, yet it is much more common than non-parasitic cysts. Such cysts are usually asymptomatic or may be found as a mass in the left upper abdomen. Ultrasonography and CT scan can detect these cysts even when asymptomatic. The best option for symptomatic cysts is splenectomy.⁹

The incidence of musculoskeletal hydatid disease is not very well established and varies between 1% and 5.4% among all cases of hydatid disease; some authors think that this low incidence may be in part due to the acidic nature of muscles because of the presence of lactic acid.^{10 11} The usual presentation is an

enlarging mass that is usually painless. Radiology, especially MRI, can establish the diagnosis. Serology again is important in suspected cases. We dealt with three cases of musculoskeletal hydatid disease which were all diagnosed preoperatively and cystectomy was performed carefully to avoid spillage of cyst contents, which did occur in one of our patients. Fortunately, we were able to remove all the spilled contents of the cyst and a thorough lavage was performed and the patient received a prophylactic dose of steroids and antihistamines.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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Learning points

- ▶ A surgeon should keep in mind that hydatid disease can occur in any part of the body from head to toe, especially in endemic areas.
- ▶ Hydatid cyst should be considered in the differential diagnosis of any cystic lesion anywhere in the body.
- ▶ A careful workup to exclude the presence of hydatid cysts in other parts of the body is critical, since secondary dissemination to such unusual sites is much more common than primary involvement.

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