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Case Report

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A wandering spleen with hydatid cyst in 16 weeks pregnancy a rare combination: A case report with review of literature

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Abstract

Wandering spleen is a rare clinical entity with less than 500 cases reported in literature, with incidence of less than 0.2%, characterised by presence of a spleen in a position other than of its normal i.e. left hypochondrium, due to hypermobility of spleen which result either from laxity or maldevelopment of its suspensory ligaments. Only few cases of wandering spleen with pregnancy have been documented in the literature. Echinococcosis (hydatid cyst) is a zoonotic infection caused by Echinococcus granulosus parasite larva. Man is an accidental intermediate host in the life cycle of parasite. We report a rare case of a wandering spleen with a hydatid cyst, presenting with recurrent abdominal pain and abdominal mass with 16 weeks pregnancy.

Keywords: Wandering Spleen, Hydatid Cyst, Pregnancy.

INTRODUCTION

Wandering spleen is a rare clinical entity with less than 500 cases reported in literature [1], with incidence of less than 0.2% [2, 3] characterised by presence of a spleen in a position other than of its normal i.e. left hypochondrium due to hypermobility of spleen, which result either from laxity or maldevelopment of its suspensory ligaments. It is 15 times more common in women than men. It occurs more commonly in women in reproductive age group (20-40 years) and usually presents as an asymptomatic abdominal or pelvic mass with acute, chronic, or intermittent abdominal pain [4]. Only few cases of wandering spleen with pregnancy have been documented in the literature [5, 6].

A wandering spleen can be divided into, congenital condition in which ligaments fail to develop properly and acquired condition like hormonal effects in pregnancy, trauma and abdominal wall laxity. However exact aetiological factors are not known [3, 7]. Wandering Spleen can result in life threatening conditions like torsion of its vascular pedicle, splenic infarction, portal hypertension and hemorrhage. Therefore, early diagnosis and treatment is required.

Echinococcosis (hydatid cyst) is a zoonotic infection caused by Echinococcus granulosus parasite larva. Man is an accidental intermediate host in the life cycle of parasite. It is an endemic disease occurring in cattle and sheep in countries of Middle East, New Zealand, India, North Africa, South America and Australia. Hydatid cyst is mainly found in Liver (60%-70%), lungs (30%); however, it can affect any organ or soft tissue [8].

Hydatid cyst in spleen is also a rare entity and constitute only 4% of all cases of abdominal hydatid cyst disease. Due to rarity of the wandering spleen along with hydatid cyst it poses a diagnostic challenge especially in non-endemic areas [8].

We report a rare case of wandering spleen with a hydatid cyst presenting with recurrent abdominal pain and abdominal mass with 16 weeks pregnancy.

CASE REPORT

A 22 years old, primigravida at 16-weeks' gestation, presented in emergency department with acute

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lower abdominal pain, more in left iliac fossa region, since one day prior, associated with two episodes of bilious vomiting. She had similar recurrent episodes of lower abdominal pain in past two months. She was attending antenatal clinic regularly with normal uneventful gestational period till then. No urinary or bowel symptoms. No history of vaginal bleed.

On examination, her abdomen was mildly tender, with a lump of size 10 cm x 8 cm firm, smooth and non-tender, mobile in left iliac fossa (Fig. 1). Vitals were stable, haematology parameters were normal. An obstetric consultation was taken.

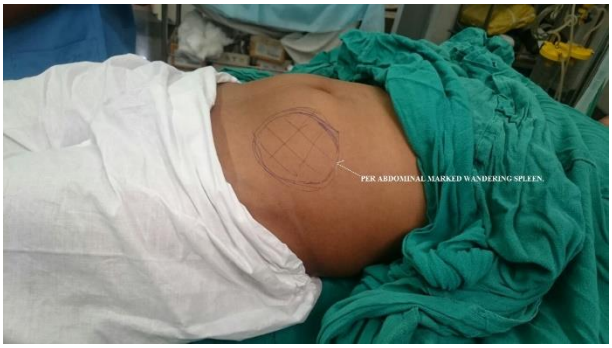

Figure 1: Showing lump of size 10cm x 8 cm in left iliac fossa.

An ultrasound scan at this point, confirmed an anteriorly lying placenta with no foetal abnormalities with absence of spleen from left hypochondrium region, which was lying in left iliac region with a simple cyst at superior pole region (Fig. 2). No other imaging was possible due to her pregnancy. She was reassured and treated symptomatically with analgesics and intravenous fluids. A diagnosis of wandering spleen with likely torsion was considered. As the patient got relieved, surgery as an option was deferred, as she was in her first trimester with a precious baby. However, she had a recurrent episode of acute abdominal pain 15 days later and was readmitted. After a repeat ultrasound and review by obstetrician, it was decided to proceed with surgery, as her rest of the blood parameters were essentially normal and she was into her early second trimester.

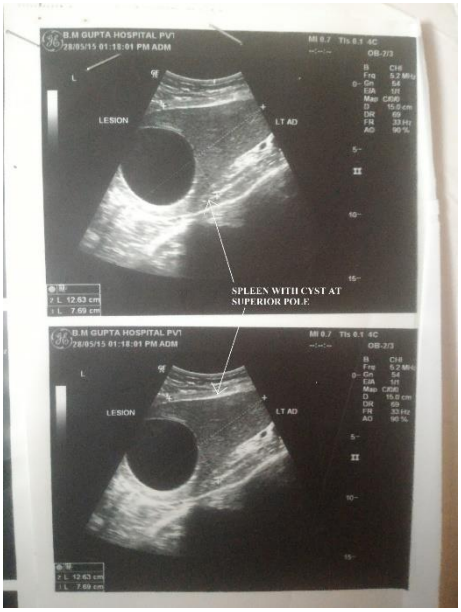

Figure 2: USG showing spleen lying in left iliac region with a simple cyst at superior pole region.

An informed consent was taken especially in regards to the risk involved to the baby. A sub-umbilical midline laparotomy was performed. A partly infarcted, hypermobile spleen was found at left iliac fossa region with long vascular pedicle. Spleen was carefully separated from colon and pancreatic tail and dissected out (Fig. 3). On examination of removed specimen, upper pole of spleen showed presence of laminated membrane with sand like fluid in it, which was later confirmed to be hydatid cyst on histopathology (Fig. 4 and 5).

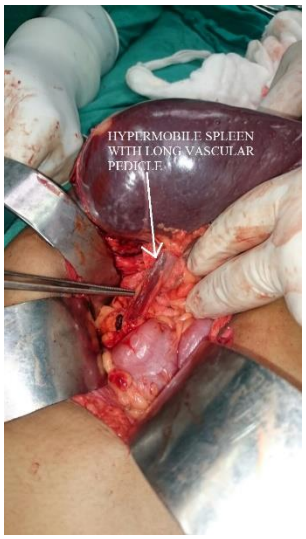

Figure 3: Spleen was carefully separated from colon and pancreatic tail and dissected out.

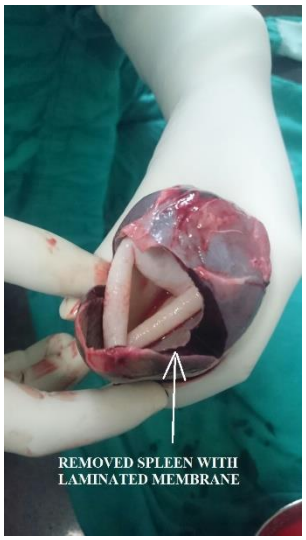

Figure 4: Cut section of spleen showing hydatid cyst.

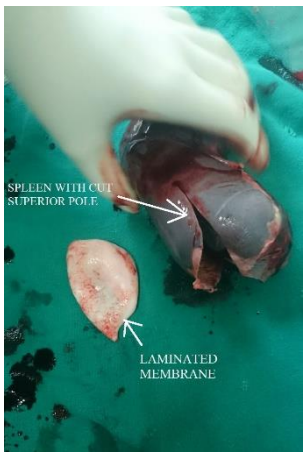

Figure 5: Cut section of spleen showing laminated membrane.

The patient made an excellent recovery post-operatively and was discharged home after taking antenatal advice. She was given Haemophilus influenzae type B, meningococcal and pneumococcal vaccinations, two weeks postoperatively. A week later, Albendazole

was started 600 mg OD. (10 mg/kg). On follow up she was asymptomatic and had a normal delivery.

DISCUSSION

"Wandering spleen", "Ectopic", "Proptotic", Floating", "Displaced", "Aberrant spleen" are the terms used to describe a rare clinical entity with less than 500 cases reported in literature [1]. Von Horne in 1667 first described wandering spleen during an autopsy [9].

Spleen develops during the 5th week of intrauterine life from mesenchymal clusters of cells within the dorsal mesogastrium. The gastrosplenic, lienorenal, phrenicocolic and splenocolic ligaments are the major supporting ligaments of spleen which are derived from dorsal mesogastrium. It is believed that non-fusion of dorsal mesogastrium with dorsal peritoneum result in abnormal position of spleen (wandering spleen) [10].

Due to rarity of this clinical entity, it is not usually considered in differential diagnosis for pelvic masses. It is more common in females as they have acquired ligamentous laxity caused by hormonal effect due to pregnancy. Wandering spleen has more incidence of occurrence in conditions which causes splenomegaly and disorders like prune belly syndrome [10].

There are multiple factors associated with etiology of wandering spleen and can be congenital or acquired. As described earlier, non-fusion of dorsal mesogastrium with dorsal peritoneum result in lack of development of major supporting ligaments of spleen which result in hypermobility of spleen. Besides pregnancy, other acquired factors include laxity of supporting ligaments as in splenomegaly, after trauma, and gastric distension [11, 12].

Patient's presentation may be variable, patient can be asymptomatic, findings can be incidental, as patient is worked up for other condition like pregnancy and when symptomatic; abdominal mass with or without pain is the most common complaint. Abdominal mass is usually painful and mobile; has painless movements towards left hypochondrium; rest of the movements are limited and painful. It has been reported in literature that 67% patients with wandering spleen had abdominal mass on presentation [12]. Abdominal pain, when chronic, is usually of dull aching in nature and when intermittent or acute, it is stabbing in nature and is due to kinking or repeated torsion of splenic vascular pedicle, resulting in venous congestion and capsular tension. De-torsion of vascular pedicle and return of blood flow may be the cause of intermittent pain and if it fails, it results in infarction of spleen causing severe abdominal pain with ischemic characteristics. Other features like fever, vomiting, pancreatitis and intestinal obstruction have also been reported in literature [12, 13]. Due to close anatomical relationship of pancreatic tail with splenic hilum pancreatitis are not uncommon.

Blood investigations are usually nonspecific and may vary in patients with hypersplenism and with pancytopenia. It is difficult to make diagnosis on clinical grounds, therefore imaging studies are necessary [7, 13]. CT scan and Ultrasonography are the imaging studies most commonly used [7]. Ultrasonography is an efficient imaging study with accuracy upto 52%, it is non-invasive and less expensive. Also, it can detect spleen in abnormal position and show absence of spleen from its normal position and when combined with Doppler it can easily evaluate the status of splenic vessels and can detect any torsion of spleen [13].

However, CT scan has been preferred over ultrasonography despite its side effect of intravenous contrast and its more cost [12]. A study done over a period of 12 years for characterizing CT findings, showed absence of spleen from its normal position to be the most common finding and same has been reported and noticed by other authors with most common site of ectopic spleen is left mid abdomen [7, 12].

The Whirl sign is a circular structure of alternating bands of radio-density and radiolucency at splenic hilum, considered to be specific sign for pedicle torsion as it is documented in all reported cases of torsion of wandering spleen [2]. Congestion appears as a stranding of fat surrounding the hilum and same has been reported in previous cases [2, 7]. Raissaki *et al* has enumerated some facts regarding the diagnosis of wandering spleen; firstly, Whirl sign should not be mistaken as intestinal intussusception; secondly, presence of left lobe in left hypochondrium may be mistaken as normal positioned spleen; and lastly, shape changed spleen can easily be confused with malignant tumors, cystic lesions or even abscesses due to congestion or infarction, therefore negative diagnosis of wandering spleen should be made after considering these facts [2].

Initially it was managed conservatively in asymptomatic patients but in view of complications, it is no more recommended. Splenectomy and splenopexy are the two procedures followed worldwide and in era of laparoscopy, laparoscopic approach is preferred unless it is contraindicated. Splenectomy is indicated in massively infarcted spleens.

Post-splenectomy sepsis is the fatal complication occurring in post-splenectomy patients and is the reason for preferring spleen preserving surgery. Encapsulated organisms, such as Streptococcus, Haemophilius influenzae and Neisseria meningitidis are the most commonly involved organisms.

To prevent post splenectomy sepsis, splenopexy is preferred wherever possible [7, 10, 12, 13]. Splenopexy can be performed in two ways, either with or without absorbable mesh. When the absorbable mesh is not used, spleen is placed in a pocket formed by mobilizing peritoneum from the underlying muscle and diaphragm and covered by peritoneum flap and sutured to stomach and colon, leaving enough space for vascular pedicle.

When mesh is used, spleen is wrapped in mesh pocket and fixed to diaphragm, abdominal wall and great curvature of stomach. Use of mesh is preferred because of difficulty in forming the peritoneal spaces by laparoscopy and use of mesh is easier and quicker.

Partial splenectomy has also been described in literature in case of partial infarction or in massive splenomegaly to make it fit to fixable size. Splenic auto-transplantation has been also described as a way of treatment [11, 13].

Echinococcus granulosus infestation most commonly affects the liver and lungs. Splenic infestation is rare because cyst embryos are filtered by the liver and lungs, with only 15% entering systemic circulation. Another way is retrograde spread from portal and splenic vessels. Secondary splenic hydatid cyst can occur after rupture of hydatid cyst in peritoneal cavity [14].

Surgery is the mainstay of the treatment for splenic hydatid cyst which can be total splenectomy or conservative like partial splenectomy, cyst eneucleation, de-roofing with omentoplasty and others. Total splenectomy is preferred, due to its lower risk of recurrence and pre-operative hemorrhage. Albendazole therapy is given post-operatively to prevent recurrence and further spread [15].

Wandering spleen, Hydatid cyst, pregnancy are clinical conditions known to us but when they occurred together, became a rare entity and after thorough search of literature, we were not able to find any case reporting three of these together as in our case.

CONCLUSION

Combination of two rare or uncommon conditions (wandering spleen with a hydatid cyst) occurs rarely, but they do occur. Our case was challenging as we had ultrasound as the only option as an imaging tool. It picked up the cyst but there was nothing to suggest that it was a

hydatid cyst. The confirmation came only when the cyst was opened post-operatively. Moreover, we had to deal with a symptomatic patient requiring surgery in late first trimester of her first pregnancy.

We feel Splenectomy was the only option due to multiple infarctions, and a hydatid cyst in one pole. We did an open procedure rather than by laparoscopy due to multiple reasons. However, this option is open to a debate.

Conflicts of interest

The authors declare no conflict of interest.

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