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Case Report:

A Rare Primary Pelvic Hydatid Cyst Presenting as Sciatica

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Abstract: Primary hydatid cyst in the pelvis is rare, and usually presents with pressure symptoms affecting the adjacent abdominal organs. We describe a rare hydatid cyst which was eroding the sacral hallow, protruding into the right sciatic foramen and presenting as a radiating pain and weakness of right lower limb due to compression of the lumbosacral nerve roots. Laparotomy with removal of cyst and postoperative treatment with albendazole is effective in controlling the disease and preventing recurrence.

Key Words: Lower limb paresis; Primary pelvic hydatid cyst; Sciatica

Introduction:

Echinococcosis is an infection caused in humans by the larval stage of the *Echinococcus granulosus* complex, These parasites are found on all continents, with areas of high prevalence in China, central Asia, the Middle East, the Mediterranean region, eastern Africa, and parts of South America. Echinococcal species have both intermediate and definitive hosts. The definitive hosts are canines that pass eggs in their feces and the intermediate hosts are sheep, cattle, humans, goats, camels, and horses for the *E. granulosus* complex. After humans ingest the eggs, embryos escape from the eggs, penetrate the intestinal mucosa, enter the portal circulation, and are carried to various organs, most commonly the liver and lungs. Larvae develop into fluid-filled unilocular hydatid cysts that consist of an external membrane and an inner germinal layer. Daughter cysts develop from the inner aspect of the germinal layer, as do germinating cystic structures called *brood capsules*. New larvae, called *protoscolices*, develop in large numbers within the brood capsule. The cysts expand slowly over a period of years.[1] Rarely, spread may occur by the lymphatic system of the bowel wall or, alternatively, by the venous circulation when the parasite has passed the liver and lungs.[2] Cysts are found in the liver (55% to 60%), lungs (30%), kidneys (2.5%), heart (2.5%), bones (2%), muscles (1%), brain (0.5%) and in other organs such as the spleen (1.5%).[3] Other rare sites include the omentum, ovaries, parametrium, pelvis[4,5], thyroid, orbit or retroperitoneum.[6] In man, infection is usually acquired in childhood. The symptoms present several years after exposure and it may take ?ve to 20 years before a diagnosis is made.

We describe a rare case of a primary pelvic hydatid cyst presenting like sciatica with right lower limb paresis. A Medline search on PubMed using the key word "primary pelvic hydatid cyst case reports" found 51 cases reported and only 5 cases have been described with a neurological deficit like sciatica.

Case Report:

A 38 years old lady presented with a radiating pain and parasthesia of right lower limb. The radiating pain started as intermittent in nature and progressively increased in intensity over a period of 3 years. Numbness and weakness involving the right lower limb had been present for 15 days. There was no associated fever, loss of appetite, backache or history of tuberculosis. The clinical examination revealed no swelling or mass in the abdomen and there was no spinal tenderness or deformity. The bimanual pelvic examination revealed a fixed mass of size 10x15x15 cms, adherent to right pelvic wall and sacrum. A normal sized uterus felt separately and bilateral parametrium, rectal mucosa appeared free. Neurological examination showed weakness of the hip extensors and abductors (4/5), the hamstrings (4/5), and the muscles of the ankle and foot (3/5) on the right side as measured on the Medical Research Council (MRC) scale. There was diminished sensation in the distribution of the L5, S1 and S2 roots on the affected side. The clinical diagnosis was that of a benign retroperitoneal pelvic tumour. Possibilities considered were a cystic neuroma arising from the sciatic nerve or the lumbosacral plexus, a sacral teratoma, or ovarian tumor or haemangioma. Ultrasonography of the abdomen revealed a large (10.7 x 15.6 cm) hypoechoic mass with echogenic septations in the presacral area posterolateral to the uterus and extending into the right sacral foramen. There was minimal right sided hydroureteronephrosis and the urinary bladder appeared normal. MRI revealed a large cystic mass in the right adnexa, presacral region with well-defined walls, situated posterior to the bladder and extending to spinal cord through right sacral foramen (Figure 1).

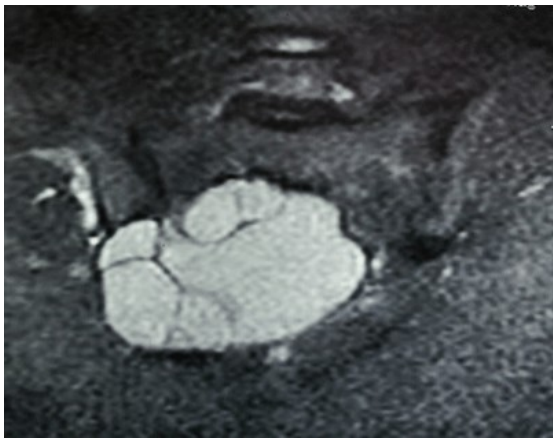

Figure 1: MRI of the pelvis showing a large cystic mass extending into the sacral foramen on the right side with well defined walls

The possible radiological diagnosis suspected was ovarian cyst, teratoma, hydatid cyst, or cystic neuroma. Aspiration of the swelling yielded approximately 15 ml of clear to straw-coloured fluid. Smear and cytospin preparation from the fluid showed acellular material with no evidence of any atypical cells or parasites. No acid-fast bacilli could be visualised in the smears. The tumor markers were within normal limits. Radiological examination of the chest was normal. We performed a laparotomy through a midline incision on the presumptive diagnosis of cystic neuroma, sacral teratoma, and hydatid cyst. A dumb-bell-shaped retroperitoneal cystic mass was found occupying and eroding the hollow of the sacrum with extending into the right sacral foramen, causing stretching of the lower lumbar and sacral nerve roots adherent to the wall of the cyst. The cyst was mobilized and it got ruptured during the process revealing multiple hydatid daughter cysts (Figure 2). The germinal layer or the endocysts were removed completely and as much of the ectocyst as possible (Figure 3). The cyst and the surgical field were washed with hypertonic saline and povidone iodine solution. The pelviabdominal organs appeared normal. Histological examination revealed the diagnosis of a hydatid cyst. The patient had considerable neurological recovery with the hip extensors and abductors and the hamstrings showing power of 5/5 on seventh postoperative day. The patient was discharged on seventh postoperative day and advised to continue per oral albendazole 400mg two times a day for 6 months.

Discussion:

The involvement of female pelvic organs by hydatid disease is extremely rare and usually not thought of until operation in the majority of reported cases. Primary pelvic hydatid disease originates in the connective tissue immediately beneath the peritoneum of the pouch of Douglas. It spreads to the uterus, ovaries, Fallopian tubes, bladder and rectum after contact. Nearly all the cases described as developing from the ovary or Fallopian tubes are really invasions from the broad ligament.[7] Our case, with a pelvic mass in right adnexal region and symptoms of nerve compression in the form of sciatica, is rare and unique. Only five such cases of primary pelvic hydatid cyst leading to a neurological deficit like sciatica are found reported on Medline search in PubMed. It is of the utmost importance that a correct preoperative diagnosis is made since all precautions must be taken to prevent dissemination and seeding of the surgical field. Deaths have been reported due to anaphylactic shock resulting from spillage during excision or biopsy after a mistaken diagnosis of a retroperitoneal tumour. In endemic regions, because of the diversity of its presentation the possibility of hydatid disease should always be borne in mind for any growing mass in the body. Diagnostic techniques such as radiography, ultrasonography, CT, MRI, and immunological tests are of value. Comparison of the Casoni and IHA tests suggests that the former is unreliable.[8,9]

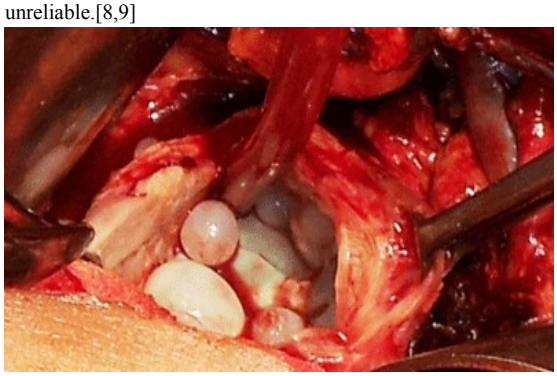

Figure 2: An Intra-operative picture of a large hydatid cyst showing multiple daughter cysts within

Figure 3: The germinal layer and multiple daughter cysts after saline wash appeared like marbles

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