Primary Sacral Hydatid Cyst Causing Cutaneous Fistula

Volkan Murat Unal1, Nail Ozdemir1, Ali Karadag1, Serdar Oguzoglu2 and Haydar Celik3

ABSTRACT

Echinococcus granulosus and Echinococcus multilocularis are the causes of hydatid disease and the main characteristic is endemic. Generally, it affects the liver and lungs. Spinal hydatidosis accounts for less than 1% of the cases. Vertebral hydatidosis is usually silent and a slowly progressive disease with a long latent period. Another rare form is the primary sacral hydatid cyst. Generally, patients suffer from back pain and neural compression symptoms. A 43-year woman was admitted with left leg pain and a fluid leakage from a cutaneous fistula on the left hip. It was diagnosed on MRI as a bilateral cystic lesion which eroded the first sacral wing, extending to the paravertebral region and left intervertebral foramen. We present a case with fluid leakage from cutaneous fistula.

Key Words: Cutaneous fistula. Hydatid cyst. Sacrum.

INTRODUCTION

The larval stages of cestodes of genus Echinococcus play a role in human hydatid cysts; and bone related hydatid cysts account for 0.5-2% of all types.1,2 Thoracic spine is 52%, and is the second most common region is lumbar vertebra 37%, sacral 5.5%, and cervical 5.5%.1,3 Vertebral hydatidosis has a silent and slowly progression which causes clinical and radiological problems.1 Portovenous anastomoses or direct extension from pulmonary focus mechanisms play a critical role in involvement of the spine.1,3 The cancellous trabeculae of vertebra may be infected by the daughter cysts.1 The vertebral hydatid cysts are described into 5 groups according to the location. These are primary intramedullary, intradural extra medullary, extra dural intraspinal, vertebral body, and paravertebral cysts.1,4 Generally, the intervertebral disc is not affected.1 Out of the related problems, spontaneous cutaneous fistulization is very rare.5,6 Surgery is successful for spinal hydatidosis affecting the bone, with surgical excision; and therapy after surgery with albendazole being the most effective.7

We present a very rare case, who had spontaneous cutaneous fistula related with primary sacral hydatid cyst.

CASE REPORT

A 43-year woman was admitted to Izmir Ataturk Research and Training Hospital with left leg pain and fluid leakage from a cutaneous fistula on the left hip. There was recurrent pain and swelling along with hemopurulent discharge stopping spontaneously with alleviation of the leg pain. She was conscious, cooperative and well oriented with no weakness. She had a ragged skin defect nearly 1x1 cm on the fistula of her hip and lower back (Figure 1A). Laboratory results were normal. Lumbosacral magnetic resonance imaging (MRI) showed multicystic lesion which eroded the first sacral wing, extending to the paravertebral region and left intervertebral foramen (Figures 2A and 2B). Computed tomography of sacrum showed some lytic lesions in the wings of the first sacral vertebra (S1) vertebrae (Figure 2C). A left hemilaminectomy was performed at L5 and S1. The lesion was located in the left wing of vertebral body at S1 level and extended to the left S1 foramen and paravertebral muscle. It was thought to be an infected cyst but the left S1 wing lesion was removed with intracavitery curettage technique (Figure 1B). Granulation tissue around the dura mater with adhesions at L5 were resected. A part of the cyst ruptured during the excision. The cavity was copiously irrigated with 3% hypertonic saline solution. Histopathological examination of the cyst wall confirmed the diagnosis of a hydatid cyst. Specific enzyme-linked...
immunosorbent assay (ELISA) and western blot analysis were positive before the surgery. Other radiological imaging showed that there were no cystic or lytic lesions in cardiothoracic vertebrae and in the other region. The patient was put on anti-helminthic (albendazole) and followed-up 3 months postoperative. She was in a stable condition and did not have any neurological deficit on first, second, sixth, and twelfth months’ examinations. A lumbosacral MRI showed a former cystic lesion, which was located on the right wing of S1 vertebra. This former right sacral wing cyst had actually become smaller. No residual or recurrent cysts in the left sacral wing.

**DISCUSSION**

Hydatid disease is a well known condition affecting liver and lungs. As to spinal hydatid cyst, it is located most commonly at the thoracic level. The other region of vertebra, and the bones of lower extremity may be infected, in descending order. The spinal hydatid cyst may either remain symptom-free for years or may cause serious complications resulting in deficit. Sacral hydatid cysts are characterized by chronicity without any clinical manifestation and are usually underdiagnosed in early stages, resulting in significant loss of bone and destruction of surrounding tissue. Generally, the first symptom is radicular back pain. Weakness of the limbs occurs in the later stage of the disease and paraplegia has been reported in around 22% of the cases. Spontaneous cutaneous fistulization is a very rare complication of hydatid disease. Cutaneous rupture, caused by spontaneous external fistula, is one of the rarest terminal lesions of hydatid cyst. Abdominal wall fistula formation has been described in acute suppurative infection. Spontaneous discharge of hydatid cyst fluid in recurrent hydatid cyst of the sacrum has been reported. The patient had a leaking cyst without any neurological deficit or pain. The anamnesis revealed that he had received surgery and antihelmintic medical therapy at L5 level hydatid cyst, and was diagnosed in 2 years following the surgery. Hydatid cyst lesions were located below the first approach within the parasacral muscles and anterior perisacral components. The authors performed excision of multiple parasacral cysts via posterior approach. In fact, the present case was quite different from the above two cases reported. This case was detected at the level of S1. There were no infiltrations in the parasacral or presacral/retroperitoneal region, or another systemic hydatid cystic lesion.

Spinal hydatid cyst should be considered in the differential diagnosis of cutaneous fistulization, at the back, pelvic, gluteal or upper thigh locations.

**REFERENCES**