Case Report

An Unusual Cause of Paraplegia: Extradural Intraspinal Hydatid Cyst

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Summary

Paraparesia, tetraplegia and radiculopathy caused by direct compression on spinal cord or ischemic changes are observed in approximately 1% of all hydatid cyst cases. A 13 year-old female patient who had been operated for lung hydatid cyst nearly 2.5 years earlier in another medical facility applied to our out-patient department with back pain, weakness in both lower extremities, paraplegia especially in the left lower extremity and paraparesia in the right lower extremity that had started 20 days earlier. During radiological examination, the reason of paraplegia was detected as extradural intraspinal hydatid cyst causing spinal cord compression and the cyst was removed surgically. The said patient, whose neurological symptoms were observed to improve during postoperative early term, is being reported here along with literature reference.

Key words: Spinal hydatid cyst, paraparesia, paraplegia, chlorhexidine gluconate

INTRODUCTION

Hydatid cyst is a disease most frequently caused by echinococcus granulosus and most commonly involves liver and lung. Spinal hydatid cyst accounts for less than 1% of all hydatid cyst diseases and is caused by primary infection of echinococcus granulosus larvae or invasion of vertebral corpus as a result of porto-vertebral venous shunts(⁶). In vertebral involvements, the thoracic spine is the most frequently involved (50% of cases) region, followed by the lumbar (20%), sacral (20%), and cervical (10%) spine. Paraplegia or root pressure has been reported in 25-84% of the patients and surgical removal of the cyst has been suggested as the most appropriate treatment option(¹).
CASE PRESENTATION

A 13 year-old female patient applied to our out-patient department with the complaints of a sudden onset low back pain, weakness in both lower extremities, paraplegia especially in the left lower extremity and paraparesia in the right lower extremity that had started 20 days earlier. The patient had no complaints related to urinary and fecal incontinence. History of the patient revealed that she had undergone a right lung surgery for lung hydatid cyst and received medical treatment for a three months term 2.5 years earlier at another medical facility. No pathology was observed in her laboratory examination. Thorax computed tomography (CT) and magnetic resonance imaging (MRI) revealed an hourglass hydatid cyst located in extradural intraspinal and paravertebral region and causing an external compression on medullary canal at T9-10 vertebrae level (Figure 1A, B).

By consulting neurosurgery, it was decided to perform a one stage surgery. Paravertebral cyst was subjected to cystectomy via right thoracotomy. Extradural intraspinal cyst was resected by vertebral corpectomy by keeping the cyst sac intact (Figure 2). The cavities of both cysts were irrigated using 0.04% chlorhexidine gluconate to prevent recurrence. Neurological complaints of the patient were observed to improve in postoperative early term and the patient started to walk too. In order to prevent recurrence, it was planned to administer albendazol treatment (10 mg/kg dose) for three months.

Figure 1: Thoracic computed tomography (A) and magnetic resonance imaging (B) showing paraspinous cystic lesion extending through the neural foramen into the epidural space and compressing the thecal sac. The thecal sac and spinal cord are displaced to the left side of the spinal canal (blue arrows).
DISCUSSION

Hydatid cyst is a life threatening parasitic disease having a slow and sneaky progress without any clinical signs but presents a 14-58% mortality risk in vertebral involvement. Vertebral hydatid cyst occurs when echinococcus larvae reaches vertebrae. Paraplegia is the most important complication of vertebral hydatid cyst. In cases of lumbar involvement, the disease is known to have a late onset compared to thoracic involvement(7). In our case, consistent with the literature, thoracic involvement had a sudden onset and the patient could not walk for the last 20 days due to paraplegia before applying to our out-patient department.

It is controversial whether spinal cord injury caused by hydatid cyst is reversible or not. However, in accordance with our clinical experience, improvement can be observed in neurological functions when compression is removed by early surgery before ischemic changes due to hydatid cyst causes irreversible nerve damage. Gürelik at al.(5) reported that paraparegia of a 52 year-old female patient improved gradually after the cyst that had caused paraparegia localized in paravertebral area in posterior mediasten was removed surgically. Likewise, it was observed in our patient that the neurological complaints which had started during the last 20 days before applying to our out-patient department improved in early postoperative term and the patient started to walk.

Echinococcosis of the musculoskeletal system is found in 0.5-4% of the patients suffering from hydatid cysts. 50% of these cases have been reported to have vertebral involvement(1,8). CT and MRI are easy to use and rapid diagnostic tools. Braithwaite and Lee(2) classified spinal hydatid cysts into 5 radiologic types: (a) intramedullary, (b) intradural extramedullary, (c) extradural intraspinal (d) vertebral, (e) paravertebral lesions extending to spinal structures. Our patient had extradural intraspinal features with an external compression on dura too. Moreover, the differential diagnoses include aneurysmal bone cysts, giant cell tumours, arachnoid cysts, solitary bone cysts, neurofibromatosis, pyogenic infections and tuberculosis(5,9).

Surgically, the right paravertebral cyst was subjected to cystectomy while the extradural intraspinal cyst was resected by corpectomy in the same session by maintaining the integrity of the cyst. During surgery, it is important to protect adjacent tissues and to resect the cyst by maintaining integrity of the cyst especially.

Figure 2: Intraoperative appearance of the cyst cavity after excision (white arrow)
when vertebra is invaded. In the literature, it has been reported that most of the cases having hydatid cysts require repeated surgery and that the recurrence rate is 50% in cases undergoing surgery.(4) Dagtekin et al. (3) reported a 22 year-old patient with hydatid cyst in whom the cysts were located between cervical paravertebral muscles. Albendazole at a dose of 10 mg/kg/day was administered 4 days before the surgical therapy and continued for 3 months. At surgery, the cyst was aspirated with removal of the cyst capsula. The patient did not have any recurrence at the end of the second year. We used intraoperative 0.04% chlorhexidine gluconate scolicidal agent in our patient, which has been proven to have a higher effectiveness compared to other agents in order to prevent recurrence in liver hydatid cysts (10). In our patient, it was known that a surgery of right hemithorax had been performed for lung hydatid cyst and she received postoperative treatment in another medical facility 2.5 years before applying to our out-patient department. However, in societies having a low socio-economic status and public awareness level and where there is no regular follow-up in terms of postoperative medication, recurrence of hydatid cysts and different metastasis areas are possible. 

**As a result**, success in treatment of hydatid cysts is only possible if cysts are totally removed via surgical principles that prevent the cyst from invading adjacent tissues. In spinal hydatid cyst disease, which may lead to a severe functional loss and mortality, patients should be followed up closely in terms of medication during postoperative term and surgery for neurological symptoms should not be delayed.

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**REFERENCES**