Spinal Hydatid disease in Iraq "endemic area"

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Abstract:

spinal hydatid disease is rare, even in endemic areas that considered hydatid disease a significant problem. Twelve cases of spinal hydatid were presented, and their management discussed. The authors reviewed most of the recent literatures and proposed a simple classification for hydatid lesions in the spine. The study stressed on consideration of this disease in the differential diagnosis of backache and spinal cord compression in endemic areas before surgical intervention so that measures can be taken to decrease complications and rate of recurrence.

Introduction

Hydatid (Greek- watery) cyst disease is very ancient, it has been mentioned in the Talmud and recognized by Hippocrates, and by the Arabian physician, Al-Rhazi who referred to hydatid liver in 900 AD. Churrier in 1807 described spinal involvement, and the first surgical intervention reported was by Reydellet in 1819(1,2).

Hydatid disease is rare but significant parasitic disease in endemic areas: Mediterranean, eastern European, east Africa, central Asia and south America, Australia and new Zealand(2,3,4).
**Pathogenesis:**

Hydatid disease is caused by the tapeworm Echinococcus, which resides in the bowel of dogs or other carnivore as a *definitive* host, segments of worm and ova pass out in the feces and later ingested by one of the *intermediate* hosts usually sheep or cattle or *man*. Here the larva are carried via the portal circulation to the liver, and occasionally beyond to other organs, where they produce cysts containing numerous scolices. Infested meat is then eaten by dogs giving rise to a new generation of tapeworm. Scolices carried in the blood stream usually settle in the liver and sometimes in the lung but occasionally in the bone producing hydatid cysts that slowly enlarge with little respect for cortical boundaries. The bones most commonly affected are the vertebrae, pelvis, femur and ribs. The cyst consist of an outer fibrous layer and an inner germinal layer containing scolices (4,5,6,7,8,9,10).

In contrast to visceral disease, bone involvement is rare accounting for less than 3% of organs involved, half of them occurring in the spine: 50% in the thoracic region, 25% in lumbar, and 25% cervical and sacral (2,3,7,9,11,12,13).

In 90% of hydatid spine, the cysts are confined to the bone and epidural spaces, while intradural cysts occur in 9% only which are either primary or secondary to previous surgery with dural tear (6,8,11,14,15,16,17,18,19,20).

In endemic areas, hydatid disease of the spine constitute a significant percentage between causes of spinal cord compression: 1.1% in Australia, 3.8% in Turkey, and 14% in Tunisia (3,8,9,12,15,21,22,23,24).

**Clinical features:**

In 20% there is a history of trauma. Usually a long history of backache &/or signs of spinal cord compression. These are gradual in onset like paraparesis and cauda equina syndrome, while rapid onset may indicate vascular compromise with less chance of recovery (6,9,13,16,20,23,25).

**Differential diagnosis:** dermoid cyst, lipoma, pyogenic and tuberculous infections, arachnoid cyst, meningocele. Bony lesions: fibrous dysplasia, enchondroma, metastatic malignancy, multiple myeloma, and giant cell tumor (1,5,7,13,24,26).

**Diagnosis:**

**Blood tests:** Eosinophelia in the peripheral blood in 20% of cases.

**Serodiagnosis:** unlike abdominal hydatid, in which ELISA (90%) and casoni (70%) tests are useful in the diagnosis, they are not in spinal hydatid because of less specificity and sensitivity (9,10,16,19,23).

**Fine needle aspiration:** should be avoided because of the risk of anaphylaxis and spillage of cyst contents if it ruptured (7,9,14,19,23,26).

**Imaging:**

**Plain x-ray:** osseous hydatid usually appears as lytic lesions surrounded by variable sclerosis, vertebral body compression fracture may be seen, paravertebral soft tissue calcification is claimed to be more suggestive of the disease. Non-osseous hydatid cyst may erode the pedicles or nearby bony surfaces (4,6,16,21,23,25).
Myelography: usually avoided because of the risk of anaphylaxis and intradural dissemination following cyst rupture(7,9,18,19).

C T scan: is the best imaging method for osseous lesions(9,13,16).

M R I: is the best imaging method for non-osseous lesions: spherical or sausage-like with tow domes, containing no septa or debries inside, and showing no enhancement on contrast injection. Post-operative MRI is useful to detect missed cysts. Scanning of head, chest and abdomen may show extra-spinal cysts(3,6,13,16,22).

Treatment:

Surgery: is indicated to confirm the diagnosis, to relief neural compression, and to maintain spine stability. In non-osseous extradural or intradural lesion, surgical removal of all cysts without rupture should result in cure(4,8,24).

Osseous lesions are the major challenge because spillage of scolices is unavoidable and total cysts removal is unachieviable,thus recurrence is the rule. The second problem is that vertebral body lesion may require extensive bony resection that necessitate grafting and stabilization usually through limited surgical field(5,9,17,19,23,25,26,27).

Complications: intraoperative death, increasing neurological deficit, misdiagnosis and cyst rupture causing spillage, anaphylaxis, CSF fistula, secondary infection, recurrence, the mortality rate: 0-3 % increasing to 15% with subsequent operations(4,9,11,15,19,20,22,23).

Scolicidal agents: the safest is 3% hypertonic saline, povidone iodine can also be used. Other agents, used in abdominal cysts, are harmful in spinal lesions like chlorhexidine, 80% alcohol, 0.5% silver nitrate, and 10% formaline(1,2,6,24,25,26).

Prognosis: theoretically, only non-osseous lesions if completely removed without spillage is curative, otherwise recurrence is the role. Up to 40% is the rate of symptoms recurrence within 2 years of initial operation, with further deterioration of neurological status and spine stability. Paraplegia is reported to occur in 45% of patients with recurrence(3,7,10,14,17,22).

Drug therapy: albendazole or better mebendazole (both are derivatives of benzimidazole) is used for 6 months in a dose of 400 mg 2 times daily. Though they are not curative, but may decrease rate of recurrence(5,8,9,11,14,21).

Material and Methods:

Al-Hilla teaching general hospital is the only surgical hospital in Al-Hilla province that deals with spine surgical diseases, it also receives cases from 7 hospitals in rural areas and sometimes cases referred from nearby provinces. A cooperation between orthopedic and neurosurgical departments started the study by collection of spinal hydatid cases in a period of about 9 years from October 1999-December 2007 …..

History, physical examination, laboratory investigations and radiographic examinations and CT &/or MRI were done for every patient presented with features of spinal cord compression. The diagnosis was unexpected in half of the cases (6/12) and predictable in 6/12 cases: 3/6 were recurrent cases, and 2 /6 were having MRI features suggestive of hydatid and
one with primary osseous lesion. Surgery performed as wide laminectomy through the posterior approach for neurological decompression at the level of spinal involvement. The surrounding surgical field was packed with mops to prevent local spillage. However, sclodical solutions, such as hypertonic saline, were not used during surgery for fear of chemical damage to the cord.

So many times, we have surprised with hydatid cyst extruded after laminectomy and some were large enough necessitating wider exposure at multiple levels. Unfortunately, cyst rupture have occurred in 2 cases, this definitely will lead to dissemination, for this reason we followed these instructions in subsequent cases:

1- putting the diagnosis in mind before surgery.
2- Careful dissection and atraumatic surgery.
3- Sealing-off surgical field.
4- Avoid cyst rupture and dural tear.

Postoperative care: albendazole 400 mg three times daily, after histological confirmation of the diagnosis, continued for 6-12 months. Gradual patient mobilization using spinal braces and physiotherapeutic care for lower limbs.

Follow up: initially every 6 weeks in the form of neurological reassessment and imaging study, then according to patient condition.

Results: (see the schedule)

Twelve patients have been seen and treated: 9 new cases plus 3 recurrent. Most of them (9/12) 75% were from rural areas. Seven were female and 5 were male with female: male ratio of 1.4:1. Their age ranged from 26-58 years with an average of 42 years. The spinal level involved was: 4 cases-dorsal, 3 cases-dorsolumbar junction, 5 cases-lumbar. All patients presented with clinical features of spinal cord compression: neurological deficit in variable grades and in different levels in the lower limbs. We have divided hydatid lesions in the spine into:

- Osseous: Primary osseous – one case
- Secondary osseous – 3 recurrent cases
- Non-osseous: Extradural – 8 cases
- Intradural – 0 cases

All patients underwent surgery; in 9/12 we did wide full laminectomy achieving decompression and cyst removal (2 cases show spillage). For the 3 recurrent cases, laminectomy at another levels was done for two, and cyst excision for one. Recurrence have been seen in 6/12 cases, 3 cases were already recurrent, 2 were those who showed spillage intraoperatively, and one with primary osseous lesion.

Discussion:

Hydatid disease is a rare but significant disease in endemic areas, about half cases of musculoskeletal hydatid involves the spine, in this study we have 12 cases of hydatid spine and this means that our area (Iraq) is endemic of hydatid disease (2,3,4).

The female: male ratio is 1.4:1 this female predilection may be due to the fact that most of farm works in Iraq is done by female rather than male.
The age range was 42 years, this late onset is related to the very slow larva growth in this intermediate stage.

The level of spinal involvement was: dorsal in 4/12 (33%); dorsolumbar in 3/12 (25%); and lumbar in 5/12(42%); this slightly differs from that of most reports: 50% dorsal; 25% lumbar; and 25% cervical and sacral (1,3,6,9,10,11,14,18,20,25).

All 12 cases are presented with neurological symptoms of the lower limbs with various grades and severity according to the level of involvement, most cases are presented with back pain (80%), radicular pain (90%), paraparesis (20%) and quadri equine (12%), these figures are nearly similar to that of other studies (4,5,8,10,12,15). Hence, the importance of remembering hydatid cyst as a possible cause of spinal cord compression especially in endemic area (3,6,9,11,23,26,27).

By reviewing most of recent literatures on spinal hydatid, we found most of them considered spinal hydatid as a part of bone disease “1% of hydatid disease patients have bone lesion, half of them affecting the spine” (1,2,4,6,7,9,10,11,13,15,17,18,19,20,23,24,25,26,27), this statement was repeated in most of these literatures and at the same time they reported that 90% of spinal hydatid lesions are nonosseous extradural.

To avoid confusion, we have classified spinal hydatid lesions into:

**Osseous**: primary osseous—affecting bony part of spine.

**Non-osseous**: primary non-osseous—either intra-dural or extra-dural.

Secondary non-osseous—follow recurrence of primary osseous lesion.

**Recurrence**:

In spite of 6-12 months albendazole therapy, recurrence have occurred in 6/12 cases within the first year of follow-up (proved by MRI), this high rate of recurrence indicates the ineffectiveness of drug therapy and the “malignant” behavior of the disease.

**Conclusion**:

Spinal hydatid disease should be considered in the differential diagnosis of spinal cord compression syndrome in endemic countries. Due to the relative rarity of the entity most diagnoses are made intraoperatively which increases the risk of future recurrences.

Extreme caution should be taken not to rupture the extradural cysts as complete removal might potentially result in cure.

Medical therapy is of no value in the treatment or control of recurrence after surgical removal with spillage of cysts.

The prognosis in cases of hydatid disease recurrence is poor in comparable to that of malignancies.

**References**:


22- Killough KK, Lusbough WB, Harkey HL. Parasitic infestations of the


### Schedule as summary of all cases

<table>
<thead>
<tr>
<th>No.</th>
<th>Sex</th>
<th>Age</th>
<th>Spinal level</th>
<th>Site</th>
<th>MR RI</th>
<th>Previous history</th>
<th>Operation</th>
<th>Recurr.</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.</td>
<td>female</td>
<td>51</td>
<td>rural</td>
<td>D9-10-11</td>
<td>Primary osseous</td>
<td>+ve</td>
<td>-ve</td>
<td>Laminectomy, cord decompression</td>
</tr>
<tr>
<td>2.</td>
<td>male</td>
<td>40</td>
<td>urban</td>
<td>L3-4</td>
<td>extradural secondary osseous</td>
<td>+ve</td>
<td>+ve</td>
<td>Laminectomy, excision of the cyst</td>
</tr>
<tr>
<td>3.</td>
<td>male</td>
<td>32</td>
<td>rural</td>
<td>L3-4</td>
<td>extradural</td>
<td>+ve</td>
<td>-ve</td>
<td>Laminectomy, excision of the cyst</td>
</tr>
<tr>
<td>4.</td>
<td>female</td>
<td>44</td>
<td>rural</td>
<td>D10-11</td>
<td>extradural</td>
<td>-ve</td>
<td>-ve</td>
<td>Laminectomy, rupture and slippage</td>
</tr>
<tr>
<td>5.</td>
<td>female</td>
<td>28</td>
<td>rural</td>
<td>D9-10</td>
<td>extradural</td>
<td>-ve</td>
<td>-ve</td>
<td>Laminectomy, excision of the cyst</td>
</tr>
<tr>
<td>6.</td>
<td>female</td>
<td>58</td>
<td>rural</td>
<td>D7-8</td>
<td>extradural</td>
<td>-ve</td>
<td>-ve</td>
<td>Laminectomy, excision of the cyst</td>
</tr>
<tr>
<td>7.</td>
<td>male</td>
<td>26</td>
<td>urban</td>
<td>D10-12-L1</td>
<td>extradural secondary osseous</td>
<td>+ve</td>
<td>+ve</td>
<td>Laminectomy, excision of the cyst</td>
</tr>
<tr>
<td>8.</td>
<td>female</td>
<td>45</td>
<td>rural</td>
<td>L4-5</td>
<td>extradural</td>
<td>-ve</td>
<td>-ve</td>
<td>Laminectomy, excision, rupture and slippage</td>
</tr>
<tr>
<td>9.</td>
<td>male</td>
<td>55</td>
<td>urban</td>
<td>D12-L1</td>
<td>extradural</td>
<td>-ve</td>
<td>-ve</td>
<td>Laminectomy, excision of the cyst</td>
</tr>
<tr>
<td>10.</td>
<td>female</td>
<td>35</td>
<td>rural</td>
<td>L4-5</td>
<td>extradural</td>
<td>-ve</td>
<td>-ve</td>
<td>Laminectomy, excision of the cyst</td>
</tr>
<tr>
<td>11.</td>
<td>female</td>
<td>48</td>
<td>rural</td>
<td>D11-12L1</td>
<td>secondary nonosseous</td>
<td>+ve</td>
<td>+ve</td>
<td>Excision of the cyst</td>
</tr>
<tr>
<td>12.</td>
<td>male</td>
<td>40</td>
<td>rural</td>
<td>L4-5</td>
<td>extradural</td>
<td>+ve</td>
<td>-ve</td>
<td>Laminectomy, excision of the cyst</td>
</tr>
</tbody>
</table>

The differences between osseous and nonosseous hydatid are briefed here:

<table>
<thead>
<tr>
<th>Incidence</th>
<th>Primary Non-osseous</th>
<th>Primary Osseous</th>
</tr>
</thead>
<tbody>
<tr>
<td>Neurological deficit</td>
<td>Early &amp; severe</td>
<td>Late &amp; less severe</td>
</tr>
<tr>
<td>Spine stability</td>
<td>Not affected</td>
<td>Affected early</td>
</tr>
<tr>
<td>Surgical approach</td>
<td>Posterior (laminectomy) &amp; cyst excision</td>
<td>Anterior (corpectomy) &amp; grafting &amp; stabilization</td>
</tr>
<tr>
<td>Prognosis</td>
<td>Cure if cyst excised without rupture</td>
<td>Recurrence is the role</td>
</tr>
</tbody>
</table>