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## Hydatid Disease of the Spine: A Survey Study from Turkey

**Summary:** Spinal hydatid disease is rare, even in rural areas where echinococcosis is endemic. Although the liver and lungs are commonly involved, spinal hydatid disease, either primary or secondary, represents an uncommon but significant manifestation of the disease. This survey study reviews 28 reports of spinal hydatid disease from Turkey during the past 5 decades. Only 14 patients also had pulmonary or some other organ infestation. The cysts affecting the spine were commonly in the thoracic region. Most patients had intraspinal extradural hydatid cysts associated with vertebral involvement. The presenting symptoms were mostly atypical, and it was interesting that most of the patients were misdiagnosed preoperatively as Pott's disease during the first decades, suggesting that new imaging techniques such as CT and MRI are the diagnostic procedures of choice for this disease. Surgery remains the best therapy for spinal hydatid disease, although adjuvant antihelminthic therapy may be necessary. There were only 15 cases of recurrence (18%); surgical intervention was palliative in all these patients and it was followed by chemotherapy. Operative mortality was very low (two patients died in the early postoperative period) and there were no complications related to treatment with antihelminthic drugs. The study indicates that hydatid disease should be considered in the differential diagnosis when radiological findings suggest spinal infections or tumors, and that surgical decompression in association with chemotherapy is the treatment of choice.

### Introduction

The type of echinococcal disease that affects the spine is caused by the parasite *Echinococcus granulosus*, a helminth belonging to the cestode group. It was alluded to by Hippocrates around 400 BC and is mentioned in the Talmud [1]. Hydatidosis, or echinococcosis, has a characteristic geographic distribution, occurring most frequently in sheep-raising regions, notably in Mediterranean countries, the Middle East, Latin America, Oceania, and South Africa [1–7]. The cysts are transported from the intestinal wall via the blood stream to the different organs. The liver and lungs are the major filters of the body, and few cysts reach any organ system. Accordingly, the cysts are most commonly located in the liver (60–70%) and lungs (10–15%); involvement of other organs is possible but rare [6, 8–13]. Echinococcosis affecting the spine is an uncommon finding. It was first described by Churrier in 1807 [14]. Hydatid disease is located in the bones in 0.5% to 2% of all cases; the vertebral column is involved in 50% of these [2, 4, 5, 7, 10, 15–21]. The treatment of choice is surgical, with removal of the cysts whenever possible. In endemic areas, preoperative diagnosis of hydatid disease is essential, because the rupture and dissemination of the cyst may result in recurrence. It is therefore still a life-threatening condition in spite of all the advances in diagnostic procedures, surgical techniques, and antimicrobial therapeutics. Turkey is one of the countries where echinococcosis is a common parasitic disease. The incidence of hydatid disease there is 50 to 390 cases per 100,000 inhabitants and it is particularly common in the rural areas [13, 22]. A correct diagnosis is important not only from the clinical point

of view, but also epidemiologically, because it permits breakdown of the epidemiological chain. In this study, a review of the literature on Turkey was conducted to determine the clinical and neuroradiological findings and treatment modalities, surgical or medical, of hydatid disease affecting the spine.

### Patients and Methods

Studies from Turkey during the past 50 years (1944–1996) which dealt with hydatid disease affecting the spine were collected. If a group published more than one paper on this subject, the preceding reports were omitted if their data were also included in the later publication. Publications involving hydatid cysts of other organs were excluded. The best of our knowledge, 28 reports in all, giving sufficient information on a total of 84 patients, were selected out of a total of 34 reports. Hydatid disease was caused by *E. granulosus* in 83 patients and *Echinococcus multilocularis* in one patient. The histories and neuroradiological and surgical findings were reviewed. Surgical findings in these 84 patients with spinal hydatid lesion were classified into three main groups described by Braithwaite and Lees [23]: 1) paraspinal hydatid disease, 2) spinal hydatid cyst, 3) intraspinal hydatid cyst (extradural, intradural, and intramedullary). Fisher's chi-square test for statistical analysis of the occurrence of recurrence was used; a probability of less than 0.05 was considered significant.

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Table 1: 84 cases of spinal hydatid disease in Turkey described in the literature.

Author	Year	No. of cases	Location
Beşe [24]	1944	1	Thoracic
Sarıbaş [11]	1948	2	Thoracic, lumbar
Sarpyener [25]	1949	1	Lumbar
Berkay [9]	1954	2	Cervical, lumbar
Kılıçhan [26]	1958	1	Lumbar
Topaloğlu [17]	1958	2	Thoracic, lumbar
Gökay and Bayülkem [27]	1959	1	Thoracic
Sanan [28]	1962	1	Thoracic
Gökay [3] <sup>a</sup>	1968	9(+1)	4 thoracic, 5 lumbar, (thoracic)
Rizeli and Çeviker [29]	1972	2	2 thoracic
Pamir et al. [2]	1984	11	7 thoracic, 4 lumbar
Özer et al. [4]	1987	5	Thoracic, 4 lumbar
Özkal et al. [5]	1987	3	2 thoracic, lumbar
Altnok et al. [6]	1989	10	2 thoracic, 2 thoracolumbar, 6 lumbar
Kılıç et al. [10] <sup>b</sup>	1989	6(+5)	6 thoracic, (thoracic, 4 lumbar)
Önder et al. [12]	1990	3	Thoracic, thoracolumbar, lumbar
Kars et al. [30]	1990	1	Cervical
Akhan et al. [19]	1991	1	Thoracic
İplikçioğlu et al. [15]	1991	1	Thoracic
Hız et al. [31]	1991	1	Lumbar
Bavbek et al. [20]	1992	1	Thoracic
Önal et al. [32]	1992	13	Not stated
Öğüt et al. [33]	1992	1	Cervical
Özer et al. [16]	1993	1	Thoracic
Tekkök et al. [7]	1993	1	Lumbar
Yeğen et al. [18]	1993	1	Sacrum
Göçer et al. [34] <sup>c</sup>	1994	1	Cervical, lumbar
Orhun et al. [35]	1996	1	Thoracic

<sup>a</sup>Because one of ten cases reported by the author had already been described by Gökay and Bayülkem [27] earlier, nine cases were considered in order to avoid an overlap; <sup>b</sup>Because five of eleven cases reported by the authors had been described by Özer et al. [4] earlier, only the remaining six cases were considered; <sup>c</sup>One patient had non-contagious lesions in the cervical and lumbar regions.

Table 2: Clinical findings in 84 patients with spinal hydatid disease.

Clinical findings	No. of cases	% of total <sup>a</sup>
Weakness of limbs	61	73
Back pain	36	43
Bowel/bladder dysfunction	27	32
Pain in the limbs	23	27
Sensory disturbance	20	24
Paravertebral swelling	2	2

<sup>a</sup>Some patients had more than one sign or symptom of spinal echinococcosis.

Table 3: Initial diagnoses of 84 patients with spinal hydatid disease.

Initial diagnosis	No. of cases	% of total
Hydatid cyst	7	8
Misdiagnoses:		
Pott's disease	14	17
Spinal tumor	13	15
Disc herniation	3	4
Not stated	47	56

**Results**

*Clinical and Neurological Results*

Sixty-one of the patients with spinal hydatid disease were male (73%) and 23 were female (27%); three out of every four patients were between 30 and 50 years old. Distribution of these patients according to year of publication is shown in Table 1 [2-7, 9-12, 15-20, 24-35]. Of the 84 patients evaluated, 22 were reported between 1944 and 1975, 19 between 1976 and 1987, and 43 between 1988 and 1996. Concurrent with infection of the spine, hydatid cysts were located in the lung or some other organ in 14 of these patients (17%). Weakness of the limbs and back pain were the most common symptoms. The presenting symptoms of the 84 patients are shown in Table 2. It is interesting that during the first 30-year period (1944-1975) 14 patients were misdiagnosed preoperatively as having Pott's disease. Initial diagnoses are given in Table 3.

*Radiological Results*

Diagnostic imaging techniques included x-rays in all cases (100%), myelography in 56 (67%), MRI in eight (10%),

CT in six (7%), and ultrasonography in one (1%). Bone involvement showed on the x-rays in the majority of the patients with spinal hydatid cyst, including lytic and cystic lesions at the vertebral body and/or lamina. In ten patients with extradural or intradural hydatid cysts only, however, routine x-rays during the pre-CT era revealed no abnormality. Except for 13 patients, all had data suggestive of the exact location of the cysts in the spine. Distribution of localization in 71 patients with spinal hydatid disease are tabulated in Table 4. Spinal echinococcosis involved the thoracic portion of the spine in 35 patients (49%), the lumbar region in 28 (39%), the cervical region in three (4%), the thoracolumbar region in three (4%), and the sacral region in one (2%). One patient had non-contagious lesions in the cervical and lumbar regions.

### Surgical Results

All but two patients were treated surgically. The surgical findings in these patients are summarized in Table 5. Forty-one individuals (58%) had predominant vertebral body involvement. Twenty-seven patients (38%) had intraspinal mass lesion causing neurological dysfunction in the absence of bone destruction. The focus of infection was localized extradurally in 18 cases and intradurally in nine. Three patients (4%) presented with purely paraspinal infection. In the remaining 19 patients, the exact location of the hydatid cyst was not stated in detail. Anthelmintics (mebendazole or albendazole) were administered postoperatively in some series. As shown in Table 6, 45 patients received chemotherapy after the operation, 38 of 85 had only surgical treatment. One who was not operated on had local radiotherapy and chemotherapy, and the other had chemotherapy alone. There were no complications that could be attributed to the prolonged administration of the drug and no side effects related to the type of anthelmintic treatment. To prevent recurrence, the cystic contents were devitalized in the majority of the cases with 3% NaCl, 10% formaldehyde, 0.5% AgNO<sub>3</sub>, and povidone iodine (Table 7).

### Follow-up Data

Follow-up information was available for 65 of the 84 patients. There were two deaths (2.4%) in hospital in the early postoperative period. The duration of the follow-up varied from 1 week to 3 years. Of the 65 patients, five had improved, seven showed no significant change, and four were worse at the last control examination. Long-term follow-up examination revealed 15 patients with recurrence (18%).

### Focus of Infection versus Recurrence Rate

The operation was considered only palliative in patients in whom the disease could not be totally eradicated because of innumerable vertebral cysts. Table 5 indicates the relationship between the focus of the infection and the recurrence rate. There was a significant difference in the recur-

Table 4: Distribution of localization in patients with spinal hydatid disease (n=71).

Localization	No. of cases	% of total
Cervical	3	4
Thoracic	35	49
Thoracolumbar	3	4
Lumbar	28	39
Sacral	1	2
Cervical and lumbar <sup>a</sup>	1	2

<sup>a</sup>One patient had two non-contagious lesions in the cervical and lumbar regions.

Table 5: Correlation of surgical findings in patients with spinal hydatid disease with recurrence rates (n=71). Fisher's exact chi-square test. P<0.05, significant.

Focus of infection	n <sup>a</sup>	Recurrence	
		No.	%
Paraspinal <sup>b</sup>	3	1	33
Spinal <sup>c</sup>	41	13	32
Intraspinal <sup>d</sup>			
extradural	17	1	6
intradural	10	-	0

<sup>a</sup>One patient treated surgically had two discrete foci of infection, one spinal and the other of intraspinal intradural origin. <sup>b</sup>Paraspinal lesions without vertebral or intraspinal involvement. <sup>c</sup>Patients with vertebral involvement typically had intraspinal extradural hydatid cyst associated with lytic and cystic lesions at corpus and/or lamina. <sup>d</sup>Intraspinal cysts producing mass effect with relative absence of bone involvement.

Table 6: Recurrence after different forms of treatment in 84 patients with spinal hydatid disease. Fisher's exact chi-square test. P<0.05, significant.

Treatment	n <sup>a</sup>	Recurrence	
		No.	%
Only surgery	38	13	34
Only chemotherapy	45	2	4
Surgery + chemotherapy	1	-	0
Radiotherapy + chemotherapy	1	-	0

<sup>a</sup>One patient had two non-contagious lesions in the cervical and lumbar regions.

rence rate between paraspinal or spinal involvement and intraspinal hydatid disease (32% and 4%, respectively) ( $p < 0.05$ ).

### Form of Treatment versus Recurrence Rate

Recurrence rates were 5% and 34% in patients treated by surgery plus chemotherapy and only surgery, respectively (Table 6). It was seen that there was a statistically significant difference in recurrence rate among patients given different forms of treatment ( $p < 0.05$ ).

Table 7: Recurrence after local application of different kinds of disinfectant in 84 patients with spinal hydatid disease. Fisher's exact chi-square test.  $P > 0.05$ , not significant.

Local disinfectant	n <sup>a, b</sup>	Recurrence	
		No.	%
3% NaCl solution	55	8	15
10% formaldehyde solution	10	4	40
0.5% AgNO <sub>3</sub> solution and povidone iodine	1	–	0
None	17	2	18

<sup>a</sup>One patient had two non-contagious lesions in the cervical and lumbar regions. <sup>b</sup>All but two patients were treated surgically with local application of disinfectant.

#### Kind of Local Disinfectant versus Recurrence Rate

Out of 82 patients treated surgically, 66 had disinfectant applied locally during the surgical procedure, including 3% NaCl solution (66%), 10% formaldehyde solution (12%), and 0.5% AgNO<sub>3</sub> solution and povidone iodine (1%) (Table 7). There was no significant difference in recurrence rates among patients treated with different kinds of local disinfectant ( $p > 0.05$ ).

#### Discussion

Although hydatid disease is rare in Europe and North America, it is a serious problem in some areas of the world including Turkey [2, 8, 13, 36]. The adult tapeworm of both unilocular and alveolar hydatid disease is found in the alimentary tract of the dog. Taxonomically, the causative organism of hydatid disease is *Echinococcus*. *E. granulosus*, causing unilocular or cystic hydatidosis in man, was responsible for spinal hydatid disease in most of the patients. In the present survey series, interestingly, *E. multilocularis* was detected in the pathologic specimen obtained by surgery in one case. Infection due to *E. multilocularis* in man is very much less common than *E. granulosus*. Dogs are infected by eating raw sheep meat containing hydatid cysts. Water, plants, and the skin of the dog may become contaminated with the eggs of tapeworms [1, 9, 37, 38]. Hydatid cysts develop in humans through the consumption of water or vegetables contaminated with ova of *E. granulosus*. The chitinous shell of the ovum, dissolved in the gastrointestinal tract by the action of pancreatic enzymes, releases six-hooklet embryos which penetrate the intestinal wall and disseminate via the portal circulation and lymphatics. Most embryos are trapped in the liver, but they may reach the right side of the heart and the lungs. Some may also reach the left side of the heart and the systemic circulation. Once the hexacanth embryo has arrived in tissue, it will form a hydatid cyst. In contrast to involvement in other tissues, osseous structures such as the vertebra constitute a friendly microenvironment for the development of echinococcal cysts [2, 3, 7, 9, 14, 15, 20, 29, 38, 39]. The level of incidence of hydatidosis varies greatly in dif-

ferent geographical regions. It is relatively frequent in agricultural and sheep-raising communities. According to World Health Organization data published in 1984, the high level of incidence with 12,000 cases per 100,000 inhabitants in Libya contrasts with the 0.14 cases per 100,000 population reported in Switzerland [40]. Thus it appears that human hydatid disease arises from close contact with parasitized dogs. In my opinion the transmission of the disease to man could be prevented by appropriate social behaviour and scrupulous hygiene.

Clinically, spinal echinococcosis manifests by radicular pain associated with objective sensory and motor disturbances and local tenderness at the level of the involved vertebrae. For this reason, it may often be confused with spinal tumor and Pott's disease in some countries, including Turkey, where tuberculosis is common [2, 9]. Spinal echinococcosis is not usually diagnosed until symptoms resulting from complications due to root and cord compression appear [2, 9, 24, 27]. In some cases, however, it is easier to make the diagnosis in endemic areas, as there is often a history of surgery and the diagnosis is confirmed by ultrasonography, CT or MRI. With the use of the best investigative techniques, it is possible to both detect the extent of the disease in soft tissue and show the viability of the hydatid cysts [7, 8, 16, 19, 20, 23, 30, 33, 34, 36]. Lytic and cystic lesions involving the vertebral body become evident on the x-ray or CT scan. However, an extradural infection producing mass effect without significant bone involvement may not be detected by these imaging techniques. Therefore, in the face of neurological deficit, the diagnostic procedure of choice is MRI. Except for eight cases investigated by MRI, both symptoms and radiological findings in the present study were atypical, indistinguishable from those of a spinal tumor or pyogenic or fungal infection of the spine. The present report shows an increase in related diagnoses from 26% in the pre-CT era to 74% in the post-CT and MRI era with the introduction of more accurate neuro-imaging. There is no doubt that CT and MRI have revolutionized neurosurgical practice for the diagnosis of hydatid cysts. They allow early diagnosis, more accurate localization than x-ray, and can show multiple lesions. The thoracic and lumbar regions are most often affected, and hydatid disease usually begins in the vertebral body [2, 9, 23, 31, 37, 38, 41, 42]. It is worthwhile to note that patients at high risk for spinal echinococcosis should be subjected to prolonged follow-up in order to diagnose and treat spinal hydatid disease at an early stage. In the present meta-analysis of the Turkish literature on spinal hydatid disease over 5 decades, most of the subjects had atypical radiological and clinical findings and only a few presented with findings typical of spinal hydatid disease. In my opinion, it is therefore a very useful compilation of rare situations in the course of hydatid disease. Primary intramedullary or intradural extramedullary localization of the hydatid cyst is exceptional. In most cases, intraspinal extradural spinal or paraspinal localization of the hydatid cysts were observed [2, 9, 19, 30, 43, 44]. Al-

though spinal cysts may coexist with pulmonary or hepatic hydatid cysts, the mechanisms of primary spinal infection by the parasite remain unclear. Intradural hydatid cysts may develop secondary to dissemination from an intracranial cyst. Prognosis is excellent after surgical intervention [14, 19, 34]. In the extradural form of the disease, however, microruptures or leakage of the echinococcal fluid during surgery result in spinal contamination and recurrence [2, 9, 18]. In accordance with these reports, 18% of the 84 patients reviewed presented with recurrent spinal disease.

To minimize the risk of recurrence, it has been advocated that preoperative administration of antihelminthics and some disinfectant solutions such as hypertonic saline, formaldehyde, povidone iodine, and AgNO<sub>3</sub> be used to devitalize viable parasites [12, 16, 18, 20]. However, the value of this preventive measure has not yet been clearly shown. On the other hand, there is agreement regarding the postoperative use of antihelminthics, such as mebendazole or albendazole, particularly for patients with multiple cysts [2, 4, 5, 8, 18, 34, 39, 45]. Based on present guidelines, it is recommended that antihelminthic chemotherapy be administered for a period of at least 3 months [1]. Although conservative treatment seems to be effective, a hydatid cyst involving the spine should be treated surgically before major complications develop [2, 15, 26–28, 32, 35, 46]. Surgery may have to be repeated several times to achieve complete eradication of echinococcosis. Pamir et al. [2] reported that 36% of their patients had previously undergone surgery for their cysts. The correct treatment of patients with spinal hydatid cysts with neurological involvement is laminectomy with extirpation of the cyst to achieve decompression. It is known that surgical decompression and total excision of the cyst, whenever possible, represents the treatment of choice but this is rarely possible [2, 3, 5, 12, 16, 18, 20, 25, 42, 47]. Cysts involving the spine cannot be safely resected or excised in toto in most patients [2, 5, 18]. An important point is that this operation should be followed by the administration of antihelminthics, as was the case in the majority of the series in the present study. As a rule, treatment of all cysts is often impossible in disseminated spinal echinococcosis. In such cases, only large symptomatic cysts should be surgically treated, antihelminthics administered continuously, and the patient should remain under close follow-up. On the other hand, some authors recommend local irradiation in association with adjuvant chemotherapy, but recurrence is inevitable [31]. In vertebral hydatidosis with compression of the spinal cord, the prognosis is poor, as this series shows.

Unfortunately, recurrence is to be expected if treatment is delayed, as in up to 40% of the cases [46]. The aim of treatment for spinal hydatid disease must be to resolve compression by laminectomy. It was recommended that disinfectant solutions be used preoperatively to avoid possible

recurrence, as shown in Table 7. However, a significant difference in recurrence in patients treated with different kinds of local disinfectant was not observed in my experience. The most important factor affecting prognosis is in my opinion the localization of the focus of infection. The possibility of recurrence must always be kept in mind especially in patients with vertebral involvement by echinococcal cysts. To differentiate recurrence from a residual cavity, it must be stressed that radiological findings alone cannot be regarded as evidence of response [1]. Based on my own experience, it is likely to be safe to observe a cyst that is asymptomatic. It is not yet established whether pre- or postoperative chemotherapy reduces recurrence under such conditions [1]. However, it seems that such therapy effectively keeps the disease asymptomatic for prolonged periods [4, 5, 18, 33]. Furthermore, it should be remembered that many of these patients were severely undernourished, and it was important to improve their general condition before surgery. There is no doubt that surgery for recurrence is associated with higher morbidity and mortality than the first surgical intervention. However, in my opinion most of these operations are only palliative and surgery may have to be repeated many times.

In summary, this retrospective survey study from Turkey provides an overview of clinical symptoms, initial diagnosis and diagnostic studies in 84 patients who had *E. granulosus* or *E. multilocularis* manifestation in the spine. It was concluded that: i) Turkey is one of the countries where echinococcosis is a common parasitic disease; ii) hydatid disease of the spine should be suspected when patients present with spinal cord compression, especially in endemic areas such as Turkey; iii) it may often be confused with Pott's disease or primary or secondary spinal tumor; iv) it usually affects the body of vertebrae but intervertebral discs are preserved; v) CT and MRI have provided additional diagnostic detailed osseous information and increased soft tissue discrimination during the past two decades; vi) spinal echinococcosis may only be intraspinal, but, in most cases, vertebral involvement by the hydatid cysts is observed; vii) when spinal involvement is present, the recurrence rate is high; viii) surgical decompression in association with chemotherapy is recommended to minimize the risk of recurrence in patients with spinal hydatid disease; ix) the localization of the focus of infection is the most important determinant of the outcome; x) chemotherapy in recurrent hydatid disease of the vertebral column should also be considered because of the increased risk of surgery; xi) it is doubtful whether the local application of disinfectant is useful to devitalize cystic contents.

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