



Original Article

Surgical outcome and prognostic factors for 39 recurrent spinal hydatid cysts

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ABSTRACT

Background: Cystic bone echinococcosis accounts for 0.5–4% of all reported cases, and 45% occur in the spine. Our aim was to review the clinical and radiological features, surgery, and outcomes for 39 patients with recurrent spinal hydatidosis.

Methods: Thirty-nine reports of patients with recurrent spinal hydatidosis (2011–2020) were followed for an average of 9.28 ± 5.60 months.

Results: Lesions occurred in descending order in the; lumbar (48.7%), thoracic (43.6%), and cervical spine (7.7%). Total cyst resection was achieved in 28 of 39 patients (71.8%). Intraoperative cyst rupture occurred in 13 patients (33.3%). The postoperative American Spinal Injury Association (ASIA) score was “good” in 82% of patients. Those with “poor” postoperative ASIA scores had a 41% incidence of recurrent thoracic hydatid cysts; further, they demonstrated significantly higher recurrence rates if cysts had ruptured intraoperatively ($P = 0.001$). In addition, laminectomy, subtotal original cyst resection ($P < 0.007$), and a thoracic location were all significantly associated with higher recurrence rates ($P < 0.04$).

Conclusion: The majority of patients demonstrated improvement following surgery for cervical hydatid cysts. Notably, those with poor outcomes typically experienced intraoperative ruptures of their thoracic hydatid cysts, contributing to high cyst recurrence rates.

Keywords: Outcome, Prognostic factors, Recurrence, Spinal hydatidosis

INTRODUCTION

Cystic echinococcosis (hydatidosis) is a parasitic disease caused by *Echinococcus granulosus*. Cystic bone echinococcosis accounts for 0.5–4% of all reported cases and 45% occur in the spine.^[10] The clinical presentation depends on the locations of cysts and includes; acute to chronic back pain, radiculopathy, motor deficits/paraparesis, sensory disturbances, and sphincter involvement. Based on magnetic resonance (MR) and/or computed tomography (CT) findings, vertebral hydatid cyst infections are subclassified into five subgroups: (Type 1) intramedullary hydatid cyst; (Type 2) intradural extramedullary hydatid cyst; (Type 3) intraspinal extradural hydatid cyst; (Type 4) vertebral hydatid cyst; and (Type 5) paravertebral hydatid cyst.^[5] On T1

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MR scans, cysts are hypointense; on T2 studies, they are homogeneously hyperintense and usually demonstrate low-intensity peripheral rim enhancement with contrast. CT scans frequently show multiple cysts, with osteolytic expansile lesions located within several vertebral bodies.^[5,10] Optimal treatment includes a laminectomy, with only a small subset warranting fusion. Postoperative additional administration of chemotherapy (i.e., albendazole) may delay recurrence, and/or reduce future complications.^[10] Here, we present the clinical, radiological, surgical, and postoperative outcomes for 39 patients undergoing surgery for hydatid spinal cysts.

MATERIALS AND METHODS

Ethics approval

This retrospective study involved anonymous data collection, and consent was not required.

Study population

We retrospectively reviewed the hospital records (clinical, radiological, and surgical) of 39 patients with spinal hydatidosis over a 10-year period (2011–2020). The study only included patients undergoing surgery for spinal hydatidosis along with reporting all patients with symptomatic recurrences warranting reoperations.

Patients’ preoperative clinical characteristics including ASIA scores

Over 10 years, 39 patients with spinal hydatidosis had surgery [Figure 1a]. Patients averaged 52.6 ± 9.04 years of age and were followed by a mean duration of 9.28 ± 5.60 months [Table 1]. On average, patients presented with 8 months of symptoms that included back pain (5.9%), radiculopathy (38.5%), paraplegia (61.5%), paraparesis (23.1%), or tetraparesis (7.7%). Patients’ preoperative and postoperative neurological status

were evaluated using the ASIA score; D and E scores were consistent with “good/excellent” outcomes, while A–C scores indicated “fair/poor” results. The preoperative ASIA Scores in 13 patients were good/excellent (ASIA D: 28.2% and ASIA E: 5.1%), while for 26 patients scores reflected more severe impairment (ASIA B: 51.3% and ASIA C: 15.4%). Interestingly, those with severe preoperative neurological impairment (ASIA B) more frequently demonstrated thoracic (28.2%) and lumbar (23.1%) spinal hydatidosis lesions, respectively [Figure 1b].

Utilization of MR/CT studies

Preoperative MR/CT studies

Patients underwent cervical, thoracic, or lumbar MR/CT studies to document the location and extent of their spinal hydatidosis. Most lesions involved the lumbar (48.7%), followed by the thoracic (43.6%) and cervical spine (7.7%). Typically, on MR, lesions consisted of multiloculated cysts extending into epidural soft tissues, while on CT scans, they presented with vertebral lysis and/or erosive bony defects. Cyst locations were most frequently type 3 (i.e., intraspinal extradural hydatid cyst in 66.7% of cases) [Table 1].

Postoperative MR studies

Postoperatively, patients underwent immediate and 1-month postoperative enhanced MR scans to confirm total resection (TR) or subtotal cyst resection (STR). Patients were additionally followed with contrast MR studies on a yearly basis for early detection of recurrent lesions.

Surgery

Patients all underwent decompressive laminectomies, supplemented selectively with instrumentation (i.e., where at least two levels were involved or with evidence of vertebral body damage and/or instability).

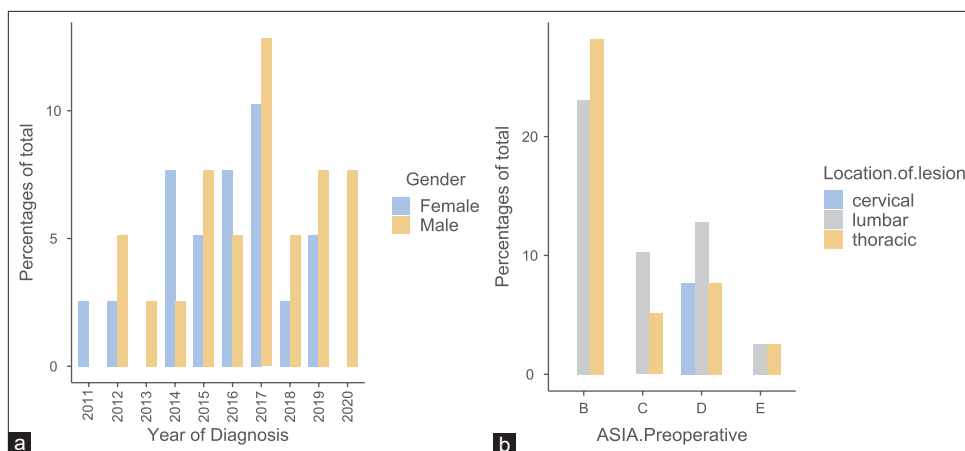


Figure 1: (a) Distribution of the cohort throughout the 10 years according to gender. (b) Clinical presentation with the American Spinal Injury Association (ASIA) score according to the location of the lesion.

Table 1: Demographic, clinical, radiological and surgical characteristics, and outcome.

Patients' characteristic	Overall (n=39)
Age of patients	
Mean (SD)	52.6±9.0 (29.0-67.0)
Gender	
Female	17 (43.6%)
Male	22 (56.4%)
Duration of symptoms	
Mean (SD)	7.7 (1.8)
Range	5.0-12.0
Length of hospital	
Mean (SD)	10.1 (9.6)
Range	2.0-60.0
Total length of follow-up	
Mean (SD)	9.3 (5.6)
Range	2.0-24.0
Symptoms	
Back pain	21 (53.9%)
Neck pain	3 (7.7%)
Radicular Pain	15 (38.5%)
Clinical presentation	
Normal	2 (5.1%)
Paraparesis	10 (25.7%)
Paraplegia	24 (61.5%)
Tetraparesis	3 (7.7%)
ASIA preoperative	
B	20 (51.3%)
C	6 (15.4%)
D	11 (28.2%)
E	2 (5.1%)
Spinal CT	
Cystic lesion+bone lysis	20 (51.3%)
Erosion+bone lysis	19 (48.7%)
Spinal MRI	
Multilocular cyst+Epidural Extension	22 (56.4%)
Multilocular cyst+Epidural Extension+SoftTissus	17 (43.6%)
Radiological Classification of spinal hydatidosis	
Type 3	26 (66.7%)
Type 4	13 (33.3%)
Location of lesion	
Cervical	3 (7.7%)
Lumbar	19 (48.7%)
Thoracic	17 (43.6%)
Surgery	
Laminectomy+STR	7 (17.9%)
Laminectomy+STR+Bone fusion	4 (10.3%)
Laminectomy+TR	27 (69.2%)
Laminectomy+TR+Bone fusion	1 (2.6%)
Intraoperative Cyst Rupture	
No	22 (56.4%)
Yes	13 (33.3%)
N/A	4 (10.3%)
ASIA postoperative	
B	4 (10.3%)
C	1 (2.6%)

(Contd...)

Table 1: Continued

Patients' characteristic	Overall (n=39)
D	21 (53.8%)
E	11 (28.2%)
N/A	2 (5.1%)
Recurrence	
No	22 (56.4%)
Yes	16 (41.0%)
N/A	1 (2.6%)

SD: Standard deviation; N/A: Not available; TR: Total cystic resection; STR: Subtotal cyst resection; ASIA: American spinal injury association.

Neurological follow-up

Neurological assessments were routinely performed preoperatively and postoperatively (i.e., last day of hospitalization, and 3, 6, 12, and 24 months postoperatively). The times between the first surgery and the first recurrence were also recorded.

Statistical analysis

Clinical and radiographic parameters were additionally analyzed using univariate analysis of preoperative and postoperative ASIA scores. Chi-square tests or Fisher's exact tests were used to analyze categorical variables, along with the *t*-test (i.e., for evaluation of continuous and ordinal variables). Survival was analyzed using the Kaplan-Meier method (i.e., to identify potential risk factors for recurrence).

RESULTS

Surgery

Following decompressive laminectomies, TR was achieved in 28 cyst cases (71.8%) and STR in 11 cases (28.2%) [Table 1]. More lumbar lesions underwent TR (41%) versus thoracic spine hydatidosis (28.2%) [Figure 2]. Only five patients required instrumented fusions (12.9%) involving the cervical, thoracic, and lumbar levels.

Intraoperative cyst rupture: A major risk factor for cyst recurrence

Nine occurred in the thoracic spine [Figure 3].

After a mean interval of 12.8 postoperative months (range: 6–24 months), 16 patients (41%) experienced cyst recurrences. In 76.9% (10/13 cases) of cases, there had been an accidental intraoperative cyst rupture during the index procedure (i.e., swabs soaked in hypertonic saline were utilized to minimize leakage in these 13 cases), while 22.7% occurred without a history of a ruptured cyst (5/22 cases) [Table 2]. Higher rates of cyst recurrence occurred in patients undergoing laminectomies for

Table 2: Risk factor of recurrence.

Parameters	Recurrence		Total (n=39)	P value
	0 (n=22)	1 (n=16)		
ASIA preoperative				
B	11.0 (50.0%)	9.0 (56.2%)	20.0 (51.3%)	
C	2.0 (9.1%)	4.0 (25.0%)	6.0 (15.4%)	
D	7.0 (31.8%)	3.0 (18.8%)	11.0 (28.2%)	
E	2.0 (9.1%)	0.0 (0.0%)	2.0 (5.1%)	
Radiological classification of spinal hydatidosis				
Type 3	17.0 (77.3%)	9.0 (56.2%)	26.0 (66.7%)	
Type 4	5.0 (22.7%)	7.0 (43.8%)	13.0 (33.3%)	
Location of lesion				0.007 ¹
Cervical	1.0 (4.5%)	2.0 (12.5%)	3.0 (7.7%)	
Lumbar	15.0 (68.2%)	3.0 (18.8%)	19.0 (48.7%)	
Thoracic	6.0 (27.3%)	11.0 (68.8%)	17.0 (43.6%)	
Surgery				<0.001 ¹
Laminectomy+STR	1.0 (4.5%)	5.0 (31.2%)	7.0 (17.9%)	
Laminectomy+STR+Bone fusion	0.0 (0.0%)	4.0 (25.0%)	4.0 (10.3%)	
Laminectomy+TR	20.0 (90.9%)	7.0 (43.8%)	27.0 (69.2%)	
Laminectomy+TR+Bone fusion	1.0 (4.5%)	0.0 (0.0%)	1.0 (2.6%)	
Intraoperative Cyst Rupture				0.001 ¹
No	17.0 (77.3%)	5.0 (31.2%)	22.0 (56.4%)	
Yes	3.0 (13.6%)	10.0 (62.5%)	13.0 (33.3%)	

N/A: Not available TR: Total cystic resection; STR: Subtotal cyst resection

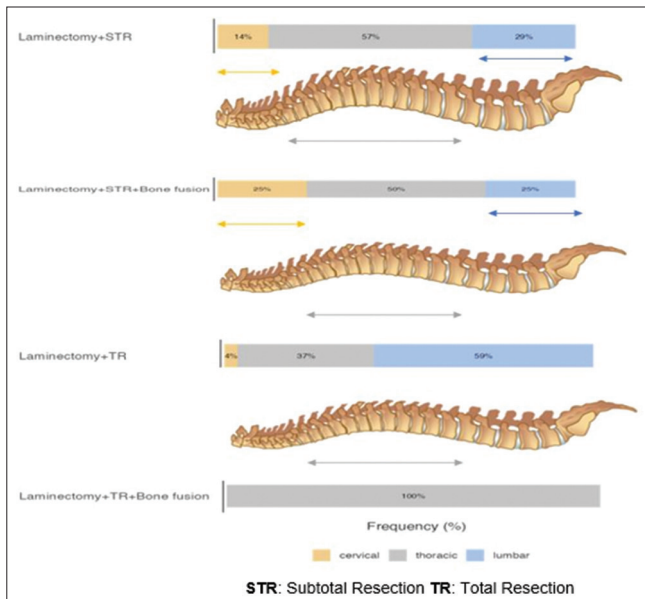


Figure 2: Types of resections according to anatomical location of the spinal hydatid cyst, STR: Subtotal resection, TR: Total resection.

thoracic lesions (2.35 times [95% CI 0.51–11; $P = 0.275$]) [Table 2 and Figure 4]. At 6-month follow-ups, a higher 65% of patients with thoracic hydatid cysts were at risk for cyst recurrence versus the lower 12-month postoperative recurrence rates of 68% for cervical and 70% for lumbar hydatid cysts.

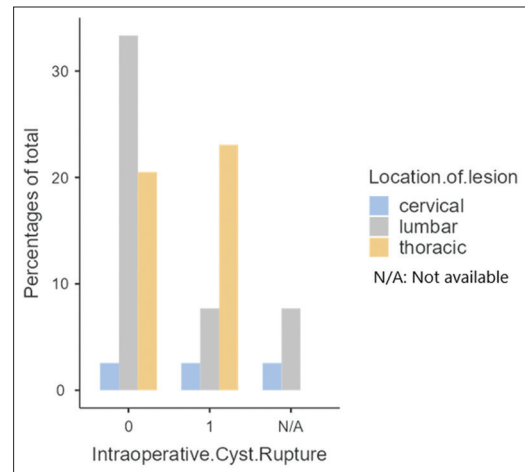


Figure 3: Distribution of intraoperative cyst rupture according to the location of the lesion.

No recurrent postoperative cerebrospinal fluid (CSF) leaks

No patients exhibited persistent postoperative Cerebrospinal fluid (CSF) leaks.

Outcomes

Postoperatively, the majority of patients were neurologically improved; the ASIA scores were “good” in 82% and “poor” in 12.9% [Figure 5 and Table 1]. Interestingly, all patients with “poor” postoperative ASIA scores had thoracic cysts.

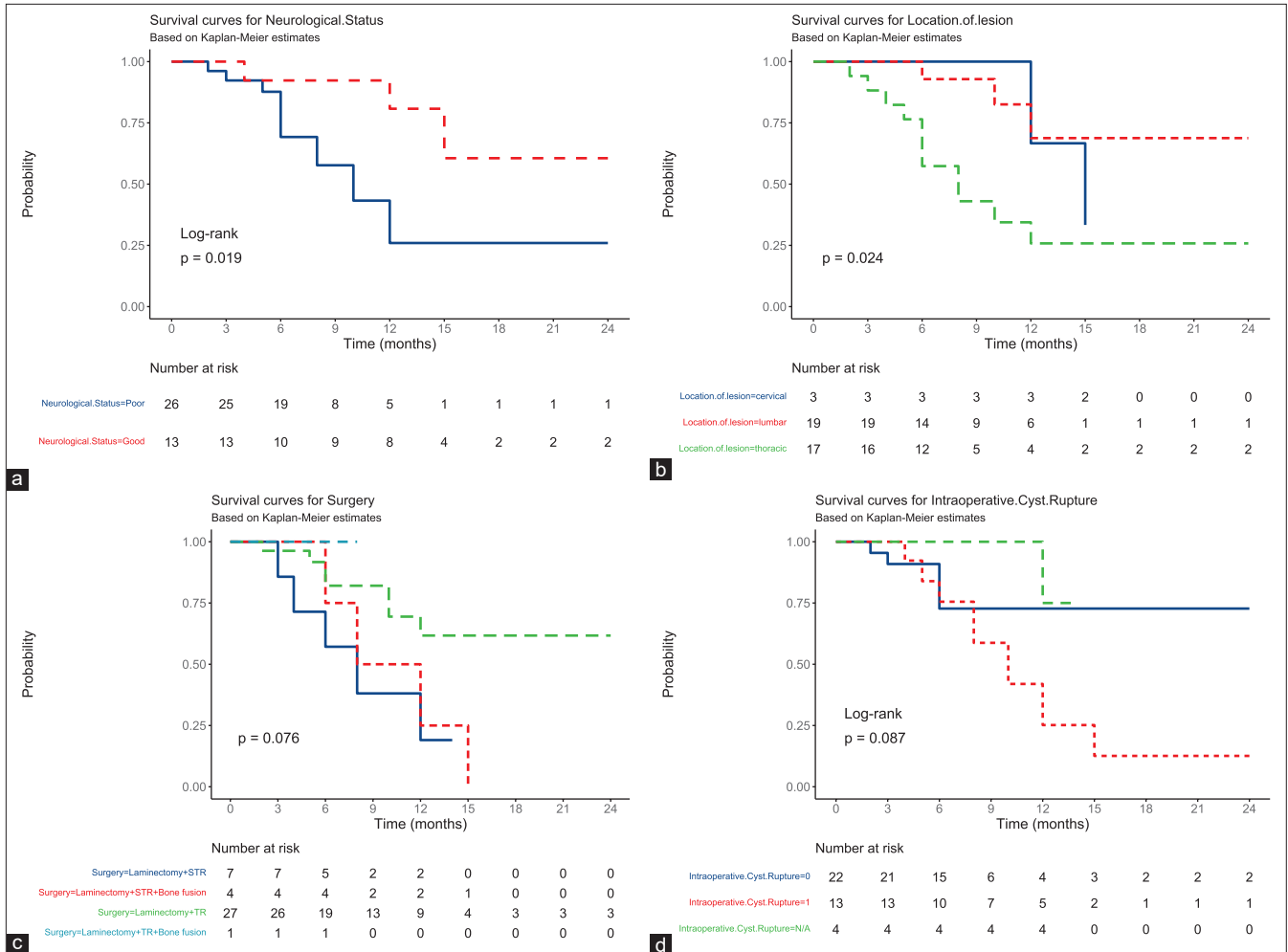


Figure 4: Multivariable cox regression analysis using Kaplan–Meier survival curves estimates risk for recurrence on: (a) neurological status, (b) location of lesion, (c) surgery, and (d) intraoperative cyst rupture.

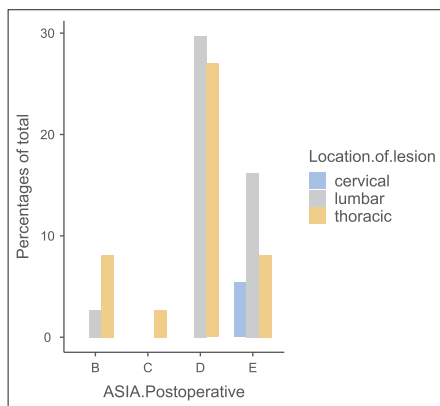


Figure 5: Distribution of postoperative American Spinal Injury Association (ASIA) score according to the location of the lesion.

DISCUSSION

Surgical management of spinal hydatid cysts

In our series of 39 patients, all underwent debridement and laminectomy to resect spinal hydatid cysts, while just five warranted additional fusions.^[1]

Recurrence rates for spinal hydatid cysts

Despite surgical advances and the use of chemotherapy, high recurrence rates remain one of the greatest challenges in treating spinal hydatid cysts.^[6] In our study, 41% of spinal hydatid cysts recurred over a mean postoperative interval of 12.8 months; this recurrence rate was comparable to those cited in other studies from the literature (i.e., range 30% to

100%).^[4-6,9] Notably, the recurrence rate was 2.35-fold higher in patients with thoracic lesions; 65% occurred within 6 postoperative months, likely reflecting a high rupture rate during thoracic cyst surgery (i.e., nine thoracic cysts ruptured intraoperatively). Within comparison, cyst recurrence rates at 12 postoperative months were 68% for cervical and 70% for lumbar hydatid cysts, respectively.

Outcomes

We found no mortalities or serious postoperative adverse events in our series. Further, we observed “good outcomes” with ASIA D-E scores in 82% of cases. Ursini *et al.* reported similar postoperative improvement in their study.^[8] Other series showed that patients largely achieved good comparable clinical outcomes.^[2,3,7] Notably, “poor outcomes” (i.e., patients with ASIA A-B-C scores) occurred exclusively in patients with thoracic cysts.

CONCLUSION

The three main factors contributing to high recurrence rates for spinal hydatidosis cysts and poor neurological outcomes included; (1) a thoracic location of the cyst, (2) subtotal rather than TR, and (3) a history of intraoperative cyst rupture during the index surgery (ASIA A-B-C).

Declaration of patient consent

Patient’s consent not required as there are no patients in this study.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The author(s) confirms that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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