

Treatment of Vertebral Hydatidosis and Factors Influencing Local Recurrence: A Systematic Review

Micaela Cinalli,¹ Gustavo Zubieta Orihuela,² Carlos A. Álvarez Martini,³ Abril Arrue,⁴ Pedro L. Bazán¹

¹Spinal Pathology Unit, Orthopedics and Traumatology Service, Hospital Interzonal General de Agudos "General San Martín", La Plata, Buenos Aires, Argentina

²Orthopedics and Traumatology Service, Hospital Obrero No. 4. Oruro, Bolivia

³Orthopedics and Traumatology Service, Hospital Municipal de Agudos "Dr. Leónidas Lucero", Buenos Aires, Argentina

ABSTRACT

Background: Bone hydatidosis is rare, but approximately half of the cases involve the spine. Treatment is challenging because of the complexity of the lesions and adjacent structures; moreover, local recurrence rates are high. The aim of this review was to describe the outcomes of surgical treatments in patients with vertebral hydatidosis and to evaluate factors associated with recurrence. **Materials and Methods:** A systematic review of articles addressing the surgical treatment of vertebral hydatidosis was performed. Data collected included characteristics of cystic lesions, type of surgery performed, and postoperative outcomes. **Results:** Data from 99 patients were analyzed. The most frequent presentation was extradural intraspinal involvement of the thoracic spine; 74.5% of patients presented with incomplete neurological deficits. Decompression surgery, resection of cysts, and spinal stabilization were performed in 75% of cases, and all patients received postoperative albendazole therapy. All patients experienced symptomatic improvement, although 27.9% did not achieve complete recovery. Among patients with follow-up longer than one year, 76% developed local recurrence. An association was found between the type of surgery performed and local recurrence ($p = 0.05$). **Conclusions:** Vertebral hydatidosis is a rare disease with slow progression and potential neurological complications. When selecting the surgical approach, lesion location, neurological involvement, and spinal stability should be considered. Recurrence is a frequent complication; however, a definitive predisposing cause could not be identified.

Keywords: Vertebral hydatidosis; surgical treatment; local recurrence.

Level of Evidence: III

Tratamiento de la hidatidosis vertebral y factores que influyen en la recidiva local. Revisión sistemática

RESUMEN

Introducción: La hidatidosis ósea es infrecuente, pero la mitad de los casos ocurre en la columna vertebral. El tratamiento es dificultoso por la complejidad de las lesiones y las estructuras adyacentes, además, las tasas de recidiva local son altas. El objetivo de esta revisión es describir los resultados de los tratamientos quirúrgicos aplicados en pacientes con hidatidosis vertebral y evaluar los factores asociados con la recidiva. **Materiales y Métodos:** Se realizó una revisión sistemática con artículos sobre el tratamiento quirúrgico de la hidatidosis vertebral. Se registró la siguiente información: características de las lesiones quísticas, tipo de cirugía realizada y resultados posoperatorios. **Resultados:** Se recolectaron datos de 99 pacientes. La lesión más frecuente fue la intraespinal extradural en la columna torácica; el 74,5% tenía déficit neurológico incompleto. En el 75% de los pacientes, se realizó una cirugía de liberación, resección de vesículas y estabilización espinal y todos recibieron tratamiento farmacológico con albendazol en el posoperatorio. En todos los pacientes, mejoraron los síntomas, aunque no de forma completa en el 27,9%. El 76% de los pacientes con un seguimiento >1 año tuvo recidiva. Se halló una asociación entre el tipo de cirugía y la recidiva local ($p = 0,05$). **Conclusiones:** La hidatidosis vertebral es una enfermedad infrecuente que tiene una progresión lenta y puede asociarse con complicaciones neurológicas. Para elegir la vía de abordaje es útil tener en cuenta la localización, la afectación neurológica y la estabilidad espinal. La recidiva es una complicación frecuente, no se puede confirmar la causa predisponente.

Palabras clave: Hidatidosis vertebral; tratamiento quirúrgico; recidiva local.

Nivel de Evidencia: III

Received on March 3rd, 2025. Accepted after evaluation on August 12th, 2025 • Dr. PEDRO L. BAZÁN • pedroluisbazan@gmail.com

 <https://orcid.org/0000-0003-0060-6558>

How to cite this article: Cinalli M, Zubieta Orihuela G, Álvarez Martini CA, Arrue A, Bazán PL. Treatment of Vertebral Hydatidosis and Factors Influencing Local Recurrence: A Systematic Review. *Rev Asoc Argent Ortop Traumatol* 2026;91(1):50-55. <https://doi.org/10.15417/issn.1852-7434.2026.91.1.2131>

INTRODUCTION

Hydatid disease is a zoonotic parasitic disease caused by *Echinococcus granulosus*. According to the World Health Organization, it is one of the most neglected and geographically widespread parasitic diseases.¹ It is more prevalent in warm regions, including South America, Mediterranean countries, the Middle East, New Zealand, central and southern Russia, Australia, China, and North and East Africa.² Humans inadvertently become intermediate hosts through contact with, or consumption of, water and food contaminated by domestic dogs.³

Hydatid cysts primarily develop in the liver and lungs (90–99.5%) and only rarely in the skeleton (0.5–4%).⁴ However, approximately half of all cases of osseous hydatid disease involve the spine, with the thoracic region being the most frequently affected (49.9%), followed by the lumbar region (21.2%).⁵ Disease progression is slow and lesions may remain inactive for prolonged periods;⁶ cyst growth rates of 1 to 5 cm per year have been reported.⁷ Diagnosis is based on imaging studies in combination with serological tests.⁴

At present, there is no consensus regarding the surgical management of this disease. The most appropriate treatment for osseous hydatid disease is en bloc resection of the affected bone; however, when spinal involvement is present, achieving this goal is often difficult because of the complexity of the lesions and adjacent anatomical structures,^{4,8} in addition to the high rates of local recurrence reported as a complication.²

Given these therapeutic challenges, the primary objective of our review was to describe postoperative outcomes in patients with vertebral hydatid disease who underwent surgical treatment. The secondary objective was to assess the association between lesion characteristics, the type of surgical procedure performed, and the occurrence of local recurrence.

MATERIALS AND METHODS

A systematic review of the literature was conducted in accordance with the PRISMA (*Preferred Reporting Items for Systematic Reviews and Meta-Analyses*) guidelines. A search was performed in the PubMed, LILACS, and SciELO databases, covering the period from January 2013 to June 2024. The search terms used were “spinal hydatidosis” and “surgical treatment.” Selection filters included human studies and publications in Spanish and English.

Articles reporting patients with a diagnosis of vertebral hydatidosis who underwent surgical treatment, with or without associated pharmacological therapy, were included.

Articles were excluded if they did not provide individual patient data, included patients who had previously received other unspecified treatments, or failed to report postoperative outcomes.

The search and initial screening of articles were conducted independently by two investigators. The second screening stage and data extraction were performed by two independent reviewer groups, with a third evaluator involved to resolve any potential disagreements.

Recorded data included study characteristics (author, year, and study design), number of patients, and demographic information.

Variables were categorized as follows:

- Characteristics of spinal involvement: spinal location of cysts and lesion type according to the Braithwaite and Lees classification⁹ (type 1, primary intramedullary cyst; type 2, intradural extramedullary cyst; type 3, extradural cyst; type 4, vertebral involvement; type 5, paravertebral cyst). Preoperative neurological status was recorded as complete deficit, incomplete deficit, or no deficit.
- Treatment performed: type of surgical procedure (group 1: curettage or cyst resection; group 2: decompression, cyst resection, and spinal stabilization; group 3: en bloc resection of the affected vertebra). Preoperative and/or postoperative pharmacological treatments administered, as well as alternative treatments, were also documented.
- Outcomes: postoperative symptoms (complete or incomplete recovery), local recurrence, and postoperative complications.

Statistical Analysis

Data were described as frequencies and percentages. Associations were assessed using the χ^2 test between cystic lesion characteristics (“spinal location” and “lesion type according to the Braithwaite and Lees classification”), type of surgery, and local recurrence.

Additionally, patients were grouped according to the surgical procedure performed, and associations between type of surgery and outcome variables were evaluated. A p value <0.05 was considered statistically significant. Statistical analyses were performed using Stata/MP version 16.0 (StataCorp LLC, College Station, Texas, USA).

RESULTS

The search of the different databases yielded 379 scientific articles. After applying the eligibility criteria, nine studies were included in the review,^{2,3,8,10-15} (Figure), representing a total of 99 patients with vertebral hydatid disease treated surgically (Table 1).

All articles consisted of case series and case reports; no studies with a higher level of evidence were identified.

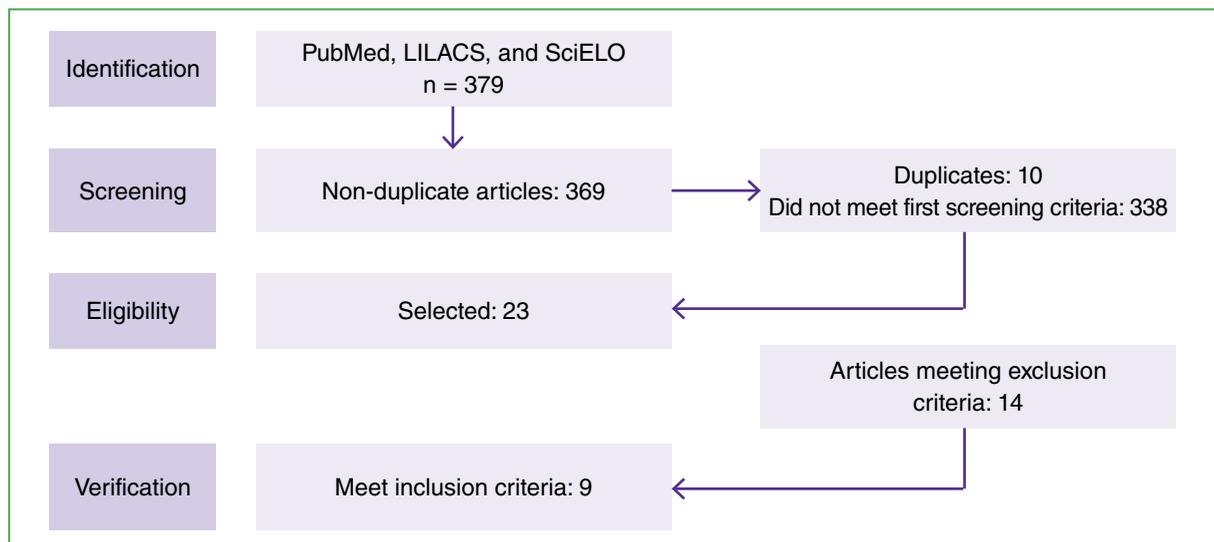


Figure. Flow chart through the different phases of a systematic review.

Table 1. Articles included in the review

Author (year)	Sample	Country	Type of study	Level of Evidence
Liu et al. ¹⁰ (2022)	1	China	Case report	IV
Luan et al. ⁸ (2022)	33	China	Case series	III
Villanueva et al. ¹¹ (2020)	2	Chile	Case report	IV
Liang et al. ³ (2019)	16	China	Case series	III
Velasco et al. ¹² (2018)	1	Uruguay	Case report	IV
Gezercan et al. ² (2017)	8	Turkey	Case series	III
El Hammoumi et al. ¹³ (2015)	1	Morocco	Case report	IV
Abdelhakim et al. ¹⁴ (2014)	1	Tunisia	Case report	IV
Kafaji et al. ¹⁵ (2013)	36	Germany	Case series	III

Lesion Characteristics

Spinal involvement was distributed as follows: thoracic spine (68 patients), lumbar spine (25 patients), cervicothoracic spine (2 patients), sacrum (2 patients), thoracolumbar spine (1 patient), and cervical spine (1 patient).

Regarding lesion type according to the Braithwaite and Lees classification, this system was not used in all articles to describe cyst location. When not specified, investigators classified the lesions based on the published imaging findings. Forty-eight patients had type 3 lesions, 32 had type 4 lesions, 17 had type 5 lesions, and 2 had type 2 lesions.

Preoperatively, 74.5% of patients presented with an incomplete neurological deficit, 1% with a complete deficit, and 25% had no neurological deficit but reported pain.

Treatment

Seventy-six patients underwent decompression surgery, cyst resection, and spinal stabilization; 17 underwent curettage or cyst resection; and the remaining six underwent en bloc resection. On average, two surgical procedures were performed per patient (range, 1–5).

Cyst resection alone was performed mainly in patients with type 4 (64%) and type 5 (35%) lesions. Decompression combined with cyst resection and stabilization was performed primarily in patients with type 3 (61%) and type 4 (21%) lesions. En bloc resection was mainly performed in patients with type 4 (83%) and type 3 (16.6%) lesions.

All patients received postoperative albendazole therapy for a mean duration of 6.5 months. One article reported preoperative treatment consisting of a single dose administered to nine patients.³

Alternative therapies included radiotherapy in four patients (total dose 6900 cGy delivered in 23 fractions over 30 days)³ and in one additional patient in whom the dose was not specified.^{3,10}

Postoperative Period

The mean follow-up duration was 4.4 years (range, 1 month to 11 years).

All patients showed clinical improvement after surgery; however, 27.9% of those with incomplete neurological deficits failed to achieve full neurological recovery.

Local recurrence was documented in 76% of patients with more than one year of follow-up.

Eight patients died during follow-up; 87.5% of these patients had type 3 lesions.

Analysis of Variables

An association was identified between the type of surgical procedure and local recurrence ($p = 0.05$). In the group that underwent decompression, curettage, and spinal stabilization, the local recurrence rate was 88.37%. It should be noted that the en bloc resection group was excluded from this analysis because of the small number of patients (Table 2).

Table 2. Relationship between type of surgery and hydatid cyst recurrence.

Type of surgery	Recurrence		Total
	No	Yes	
Curettage/cyst resection	10	5	15
Decompression, resection, and spinal stabilization	24	38	62
Total	34	43	77

Pearson's $\chi^2 = 3.82$; $p = 0.05$.

No association was found between lesion level and local recurrence ($p = 0.49$), nor between lesion type (according to the Braithwaite and Lees classification) and local recurrence ($p = 0.48$).

DISCUSSION

Vertebral hydatid disease is an uncommon condition with slow progression, but it may lead to neurological complications.¹⁶ In our search, only nine articles published over the past 10 years reported patients with spinal hydatid disease treated surgically.

The thoracic spine was the most frequently affected region, consistent with previously published data.⁵ In most articles, the Braithwaite and Lees classification⁹ was used to describe cyst location. Although this classification is purely descriptive, we believe it may be useful for planning the surgical approach and technique.

At present, there is no expert consensus regarding the management of osseous hydatid disease. Radical surgery has been proposed as a curative option,^{5,6} but complete removal of spinal cysts is often difficult to achieve, compounded by the risk of complications due to their proximity to neural structures. When radical treatment is not feasible, palliative surgery combined with long-term pharmacological therapy may be considered. The most commonly used agent is albendazole, administered at a recommended dose of 10–15 mg/kg/day for at least 6 consecutive months, to improve prognosis and reduce recurrence rates.^{4,5}

Local recurrence is the most frequent postoperative complication. Cyst rupture during surgery has been suggested as a contributing factor;¹⁷ however, this information was not explicitly reported in all articles, precluding comparative analysis of this variable. Certain strategies, such as irrigation with hypertonic saline solution, have been recommended to reduce recurrence rates.⁵ Only a small number of patients received conventional radiotherapy, but there is no evidence in the literature supporting its effectiveness.^{5,17}

An association was observed between the type of surgery and local recurrence, with higher complication rates in more aggressive procedures involving decompression and spinal stabilization. Nevertheless, given the level of evidence of the included studies, unreported factors, such as intraoperative cyst rupture or the use of saline irrigation, may have influenced these results.

CONCLUSIONS

The optimal choice of surgical technique for the treatment of vertebral hydatid disease remains unclear in the literature. Consideration of cyst location, neurological involvement, and spinal stability may be helpful when deciding between radical surgery and palliative procedures combined with pharmacological treatment.

Local recurrence is a very common complication, particularly following more complex surgical procedures; however, it is not possible to determine with certainty which factors most strongly influence its occurrence.

Conflict of interest: The authors declare no conflicts of interest.

M. Cinalli ORCID ID: <https://orcid.org/0000-0003-2057-4469>

G. Zubieta Orihuela ORCID ID: <https://orcid.org/0009-0003-7588-8577>

C. A. Álvarez Martini ORCID ID: <https://orcid.org/0009-0000-6160-9168>

A. Arrue ORCID ID: <https://orcid.org/0009-0003-5872-4634>

REFERENCES

1. WHO. *World Health Statistics 2015*. Geneva: World Health Organization; 2015, p. 162.
2. Gezercan Y, Ökten AI, Çavuş G, Açık V, Bilgin E. Spinal hydatid cyst disease. *World Neurosurg* 2017;108:407-17. <https://doi.org/10.1016/j.wneu.2017.09.015>
3. Liang Q, Xiang H, Xu L, Wen H, Tian Z, Yunus A, et al. Treatment experiences of thoracic spinal hydatidosis: a single-center case-series study. *Int J Infect Dis* 2019; 89:163-8. <https://doi.org/10.1016/j.ijid.2019.09.024>
4. Meng Y, Ren Q, Xiao J, Sun H, Huang Y, Liu Y, et al. Progress of research on the diagnosis and treatment of bone cystic echinococcosis. *Front Microbiol* 2023;14:1273870. <https://doi.org/10.3389/fmicb.2023.1273870>
5. Neumayr A, Tamarozzi F, Goblirsch S, Blum J, Brunetti E. Spinal cystic echinococcosis--a systematic analysis and review of the literature: part 1. Epidemiology and anatomy. *PLoS Negl Trop Dis* 2013;7(9):e2450. <https://doi.org/10.1371/journal.pntd.0002450>

6. Cattaneo L, Manciuilli T, Cretu CM, Giordani MT, Angheben A, Bartoloni A, et al. Cystic echinococcosis of the bone: A European Multicenter Study. *Am J Trop Med Hyg* 2019;100(3):617-21. <https://doi.org/10.4269/ajtmh.18-0758>
7. Kaloostian PE, Gokaslan ZL. Spinal hydatid disease: a multidisciplinary pathology. *World Neurosurg* 2015;83(1):52-3. <https://doi.org/10.1016/j.wneu.2013.07.106>
8. Luan H, Liu K, Deng Q, Sheng W, Maimaiti M, Guo H, et al. Multiple debridement of cavity lesions combined with antiparasitic chemotherapy in the treatment of mid or advanced spinal echinococcosis: a retrospective study of 33 patients. *Int J Infect Dis* 2022;114:261-7. <https://doi.org/10.1016/j.ijid.2021.11.014>
9. Braithwaite PA, Lees RF. Vertebral hydatid disease: radiological assessment. *Radiology* 1981;140(3):763-6. <https://doi.org/10.1148/radiology.140.3.7280247>
10. Liu P, Feng H, Liu J. A case of extensive thoracolumbar spinal intradural cystic echinococcosis. *World Neurosurg* 2022;165:89-90. <https://doi.org/10.1016/j.wneu.2022.06.034>
11. Villanueva FJS, Barbieri SL, Gallardo FA. Hidatidosis vertebral intrarraquídea. Una patología médico-quirúrgica. Reporte de dos casos. *Rev Asoc Argent Ortop Traumatol* 2020;85(1):56-64. <https://doi.org/10.15417/issn.1852-7434.2020.85.1874>
12. Velasco JM, Sapriza S, Galli N, García F. Vertebral hydatidosis: bibliographical review and clinical case report. *Coluna/Columna* 2018;17(4):326-9. <https://doi.org/10.1590/S1808-185120181704191824>
13. El Hammoumi MM, El Mostarchid B, Kabiri EH. Posterior approach to intrathoracic hydatid cyst invading the dorsal spine. *Arch Bronconeumol* 2015;51(11):600-1. <https://doi.org/10.1016/j.arbres.2015.05.011>
14. Abdelhakim K, Khalil A, Haroune B, Oubaid M, Mondher M. A case of sacral hydatid cyst. *Int J Surg Case Rep* 2014;5(7):434-6. <https://doi.org/10.1016/j.ijscr.2014.03.025>
15. Kafaji A, Al-Zain T, Lemcke J, Al-Zain F. Spinal manifestation of hydatid disease: a case series of 36 patients. *World Neurosurg* 2013;80(5):620-6. <https://doi.org/10.1016/j.wneu.2013.06.013>
16. Sioutis S, Reppas L, Bekos A, Soulioti E, Saranteas T, Koulalis D, et al. Echinococcosis of the spine. *EFORT Open Rev* 2021;6(4):288-96. <https://doi.org/10.1302/2058-5241.6.200130>
17. Neumayr A, Tamarozzi F, Goblirsch S, Blum J, Brunetti E. Spinal cystic echinococcosis--a systematic analysis and review of the literature: part 2. Treatment, follow-up and outcome. *PLoS Negl Trop Dis* 2013;7(9):e2458. <https://doi.org/10.1371/journal.pntd.0002458>