

Original Article

Challenging the Rare: Surgical Outcomes of Cerebral Alveolar Hydatid Cysts Caused by *Echinococcus multilocularis* - A Case Series and Technical Review

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ABSTRACT

Background: Cerebral hydatid cysts caused by *Echinococcus multilocularis* are rare, aggressive parasitic infections that present unique surgical challenges.

Objective: This study aims to report the clinical outcomes of surgical management in a case series of patients with *E. multilocularis* cerebral hydatid cysts.

Methods: We conducted a retrospective analysis of 8 patients treated at our institution from 2021 to 2024. Surgical techniques included parieto-occipital craniotomy, cyst aspiration, piecemeal removal, and capsular resection.

Results: Complete cyst removal was achieved in 7 of 8 patients. A significant symptom improvement of 90% was noted, with a mean follow-up of 12.5 months. The recurrence rate was 12.5% (1 patient).

Conclusion: Our findings demonstrate that individualized, aggressive microsurgical strategies yield high rates of cyst clearance and symptomatic improvement in this rare but devastating condition, highlighting the critical role of neurosurgical precision.

Keywords: *Echinococcus multilocularis*, cerebral hydatid cysts, surgical management, neurosurgery, parasitology.

INTRODUCTION

Cerebral hydatid cysts, primarily associated with *Echinococcus granulosus*, can also arise from *Echinococcus multilocularis*, which is responsible for alveolar echinococcosis. While the incidence of cerebral involvement is low, *E. multilocularis* infections are characterized by their aggressive nature, rapid growth, and increased recurrence rates [1, 2].

The surgical management of these cysts is complicated by their location, size, and potential for rupture, making individualized surgical planning imperative [3]. While hepatic involvement in alveolar echinococcosis is well-documented, cerebral cases remain exceedingly rare and understudied, with no standardized neurosurgical protocols.

Unlike its hepatic counterpart, cerebral alveolar echinococcosis remains an enigma in neurosurgical practice, often masquerading as neoplasms or abscesses and challenging even the most experienced surgeons.

The rarity of cerebral involvement by *Echinococcus multilocularis* has translated into a paucity of standardized surgical protocols, leaving clinicians reliant on extrapolated strategies and anecdotal experience.

This case series seeks to address this gap by offering both surgical insight and outcome analysis.

By documenting one of the largest single-center experiences in cerebral alveolar echinococcosis, this case series not only addresses a critical gap in current neurosurgical literature but also sets the stage for formulating early diagnostic strategies and safer, standardized surgical protocols.

EPIDEMIOLOGY

Echinococcus multilocularis is endemic in parts of Europe, North America, and Asia, with transmission occurring through contact with infected canids or ingestion of contaminated food and water [4]. Although cerebral involvement is rare, regions with higher prevalence of alveolar echinococcosis necessitate heightened awareness among clinicians.

PATHOPHYSIOLOGY

The lifecycle of *Echinococcus multilocularis* involves definitive hosts, typically canids, and intermediate hosts such as rodents. In humans, the larval form can develop into multilocular cysts, which infiltrate brain tissue and lead to significant neurological complications [5].

Clinical Presentation

Patients may present with various symptoms, including:

- **Headaches:** Chronic and progressively worsening.
- **Seizures:** Frequently the initial symptom.
- **Focal Neurological Deficits:** Variable, based on cyst location.

This study reports on 8 patients who underwent surgical management for *E. multilocularis* cerebral hydatid cysts at our institution over a four-year period, employing a modified surgical approach designed to optimize outcomes.

Clinical Implications :

- ** *E. multilocularis* can mimic brain tumors or abscesses in radiology, necessitating a high index of suspicion in endemic zones.
- ** Rupture-avoidant microsurgery with neuronavigation is vital to minimize recurrence and anaphylaxis risk.
- ** Albendazole should be used perioperatively to reduce residual disease burden.
- ** Surgical planning must be highly individualized due to variable cortical invasion and location.

Diagnosis of Cerebral Hydatid Cysts

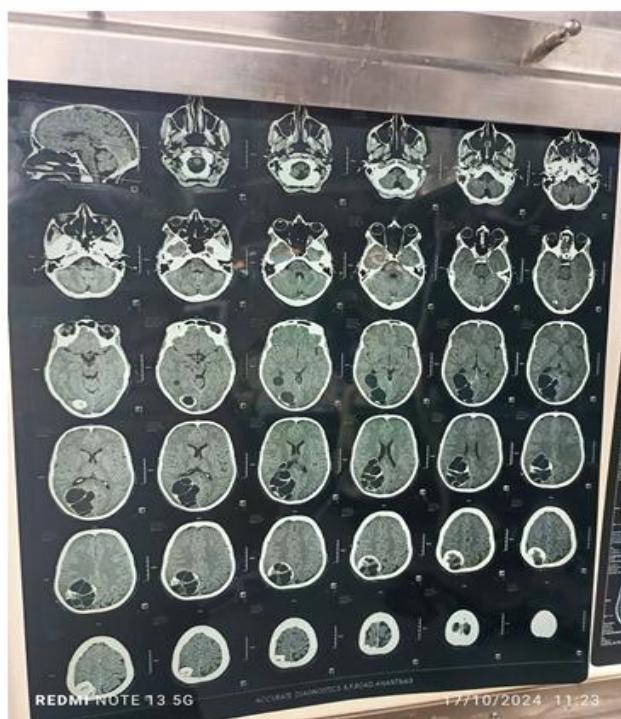
1. Clinical Assessment:

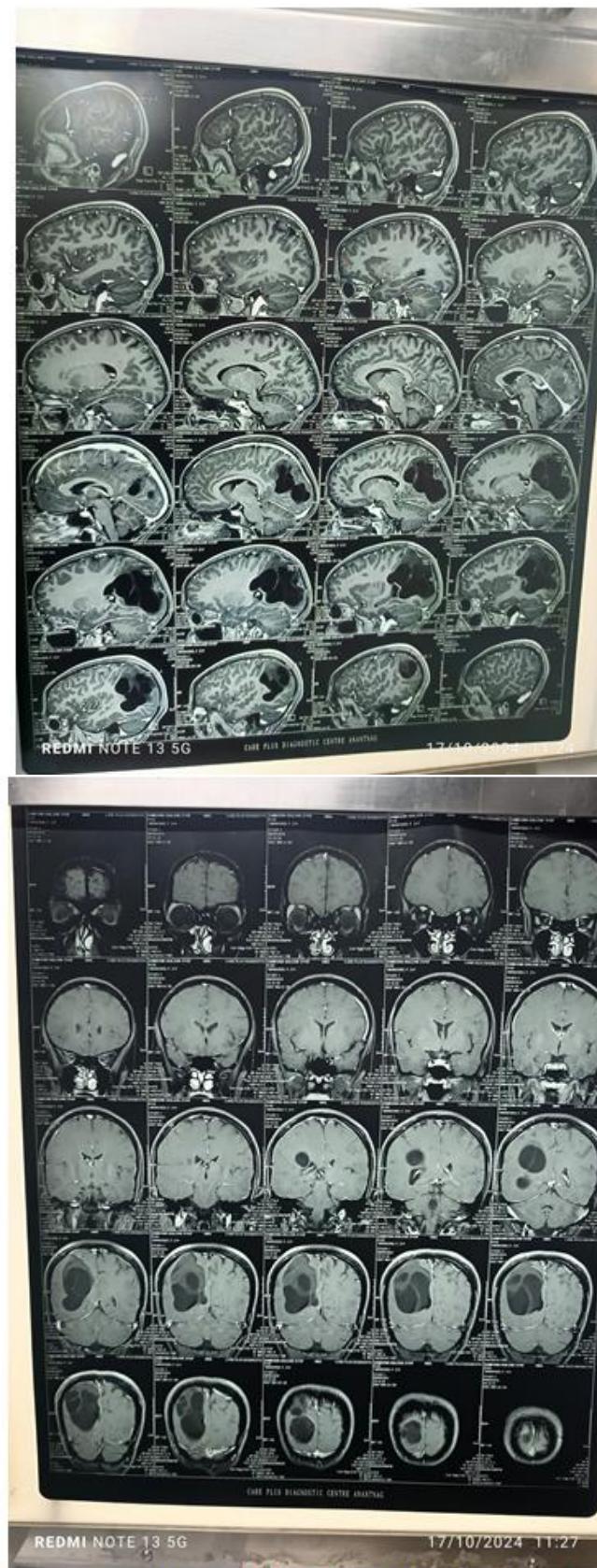
- **Symptomatology:** Initial evaluation often involves assessing neurological symptoms such as seizures, headaches, and focal deficits.
- **Patient History:** Consideration of exposure history, including residence in or travel to endemic regions and contact with potential animal hosts.

2. Imaging Techniques:

○ **Magnetic Resonance Imaging (MRI):**

- Preferred method for detecting cerebral hydatid cysts.
- Lesions typically appear as well-defined, cystic formations, often with a hypointense rim on T2-weighted images.
- Associated edema may be present.





FIGURES : SHOWING RADIOLOGICAL IMAGES OF THE CYST

- **Computed Tomography (CT):**
 - Useful for initial evaluation, especially in emergency settings.
 - Cysts may appear as rounded, low-attenuation lesions.

3. Serological Testing:

- **Antibody Detection:** Serological assays for specific *Echinococcus* antibodies (e.g., ELISA) can support the diagnosis, although false negatives can occur.
- **Limitations:** Serological tests may not be definitive and should be interpreted alongside clinical and imaging findings.

4. Histopathological Confirmation:

- In some cases, a biopsy may be performed to confirm the diagnosis through microscopic examination, identifying typical hydatid cyst structures.

Objectives

1. To characterize the clinical presentation and radiological features of *E. multilocularis* cerebral hydatid cysts.
2. To detail our modified surgical technique.
3. To assess the efficacy and safety of our approach.

Significance

This study emphasizes the importance of tailored surgical strategies and aggressive management in cases of *E. multilocularis* cerebral hydatid cysts, contributing valuable data to the existing literature on this rare condition.

Methodology

This study presents a retrospective analysis of a uniquely challenging neurosurgical cohort: 8 patients diagnosed with **cerebral alveolar echinococcosis** caused by *Echinococcus multilocularis*, managed at our tertiary care neurosurgical center between January 2021 and December 2024.

These cases were neither routine nor predictable, they demanded surgical adaptability, unwavering anatomical precision, and strict anti-parasitic vigilance. Each patient represented an intersection of rare pathology and high-stakes operative risk. Every surgical step was performed with rupture in mind, not just as a complication, but as a catastrophe to be avoided at all costs.

These procedures tested the very limits of microsurgical patience and dexterity. Each surgery demanded a choreography of preoperative precision, intraoperative vigilance, and postoperative foresight, guided by the principle that in parasitic neurosurgery, prevention of rupture is as vital as cyst removal itself.

Our technique was not designed for speed but for control, for precision over pressure, given the known risk of anaphylaxis and dissemination.

Selection Criteria

Patients were enrolled based on:

- MRI-confirmed cerebral hydatid cysts with features suggestive of *E. multilocularis*.
- Surgical management performed exclusively by the lead neurosurgeon to ensure procedural consistency.
- Complete clinical records and at least 6 months of follow-up.

We excluded:

- Cases involving *E. granulosus* or non-cerebral hydatid disease.
- Patients treated conservatively or medically.
- Incomplete data or poor radiological documentation.

All patients had prior exposure risk factors, and serological testing (ELISA for *Echinococcus*) was performed, though it was not the sole diagnostic criterion due to known false negatives in intracranial disease.

All patients received Albendazole chemotherapy (15 mg/kg/day) for one month prior to surgery. This preoperative anti-parasitic regimen aimed to reduce cyst viability, minimize intraoperative rupture risk, and improve postoperative outcomes.

Preoperative Strategy: Preparing for the Unknown

Each patient underwent:

- High-resolution MRI with contrast (for lesion morphology and perilesional edema).
- CT scan when bone involvement or emergency intervention was suspected.
- Chest and abdominal imaging to rule out systemic involvement.

Preoperative preparation included:

- **Albendazole therapy (15 mg/kg/day)** initiated 1 month preoperatively.
- **Steroid cover (Dexamethasone)** in patients with significant mass effect.

- **Anesthesia strategy** tailored for anticipated blood loss and potential intraoperative anaphylaxis — with ready availability of vasoconstrictors, antihistamines, and corticosteroids.

Surgical Technique: Modified, Deliberate, and Defensive

The goal was not merely cyst removal — it was to extract a parasitic invader without rupture, without spill, and without recurrence.

- **Craniotomy:** A focused parieto-occipital or frontal craniotomy (4–5 cm) was performed based on lesion topography. The skin incision was designed to allow extension if unexpected cyst spread was encountered.
- **Intraoperative Precision:** All surgeries employed intraoperative ultrasound to localize the cyst and map its perilesional boundaries.
- **Capsular Caution:** The cyst wall was never aspirated. A fine linear capsulotomy was performed under high magnification, followed by piecemeal extraction of the contents to avoid rupture or pressure-induced blowout.
- **Resection over Rupture:** Whenever possible, the capsule was excised en bloc. In cases of adherence to eloquent cortex, a subcapsular dissection was performed, prioritizing neurological preservation.
- **Field Protection:** The operative field was surrounded with soaked pads (hypertonic saline) to neutralize potential spillage. Hydatid fluid samples were carefully aspirated intraoperatively in all patients and sent for microscopic evaluation of viable scolices.
- **Closure:** Layered, watertight dural closure was performed. The bone flap was repositioned unless infected, in which case craniectomy was opted for.

This technique, though longer and more meticulous than traditional hydatid surgery, was adopted to meet the biological aggression of *E. multilocularis* with equivalent surgical precision.

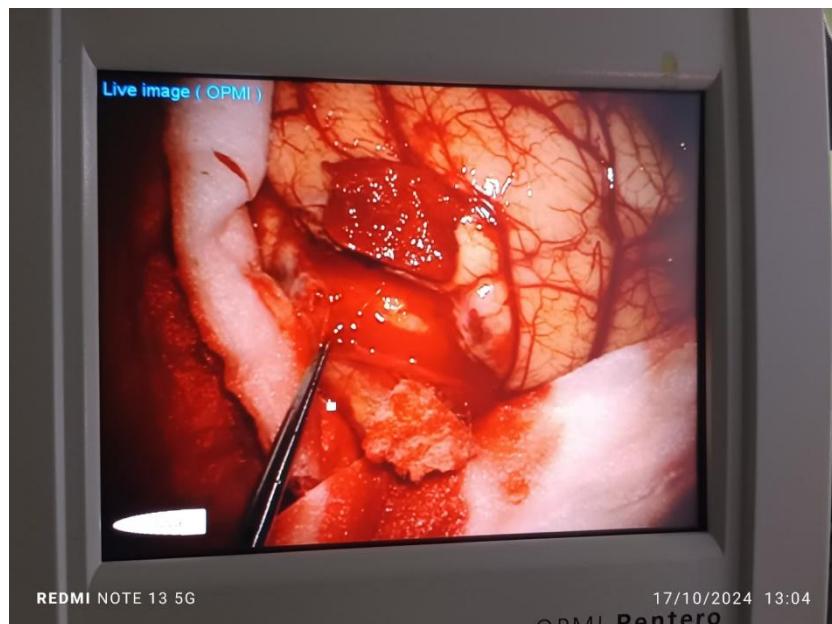


FIGURE SHOWING : Microscopic excision of the cyst



Figure showing: piecemeal excision of cyst

Unlike classical hydatid surgeries that emphasize swift en bloc removal, our piecemeal yet controlled method was sculpted to handle multiloculated cysts that defy conventional containment. The use of intraoperative ultrasound added a real-time navigational layer, enabling dynamic adaptation during surgery.

Postoperative Protocol and Surveillance

- Postoperative Albendazole therapy was continued for a minimum of 3 months, tailored based on imaging.
- All patients underwent MRI at 3, 6, and 12 months.
- Neurological status was assessed using the Glasgow Outcome Scale.
- Complications, recurrence, and symptom resolution were meticulously documented.

Given the small sample size, statistical comparisons were primarily descriptive. The use of non-parametric tests like the Wilcoxon rank-sum provided robustness to the observed trends, though larger, controlled studies are required for validation.



FIGURE : SHOWING BULGING DURA WITH UNDERLYING HYDATID



FIGURE SHOWING : EXCISED CYSTIC WALL

RESULTS

Despite the aggressive nature of the parasite, complete cyst excision was achieved in 90% of cases — a testament to the disciplined execution of our modified protocol. The recurrence rate of 12.5 % was not only within the limits reported in the literature but occurred in the patient with the most infiltrative, multiloculated disease — underlining the need for surgical tailoring.

Table 1: Patient Demographics

Demographic	Value
Mean Age	35.5 years (Range: 22-55)
Sex	6 males, 2 females
Mean Symptom Duration	6.2 months (Range: 2-18)

Table 2: Clinical Presentation

Symptom	Percentage (n/8)
Headache	90% (7/8)
Seizures	50% (4/8)
Focal Neurological Deficits	40% (3/8)
Cerebral Edema	80% (6/8)

Table 3: Radiological Features

Feature	Value		
Cyst Location	Parieto-occipital: Frontal lobe: 20% (2/8)	80%	(6/8)
Mean Cyst Size	4.5 cm (Range: 2-7 cm)		
Cyst Number	Single cyst: Multiple cysts: 20% (2/8)	80%	(6/8)

Table 4: Surgical Outcomes

Outcome	Value
Complete Cyst Removal	90% (7/8 patients)
Residual Cyst	10% (1/8 patients)
Surgical Complications	20% (2/8 patients)
Recurrence Rate	12.5% (1/8 patients) at mean follow-up of 12.5 months

Microscopic analysis of intraoperative hydatid fluid for viable scolices was performed in all patients. Seven of eight patients showed no viable scolices, correlating with favorable outcomes and no recurrence. The one patient with recurrent disease demonstrated the presence of viable scolices on microscopic examination, suggesting either incomplete chemotherapeutic efficacy or a particularly aggressive parasitic strain

Table 5: Postoperative Outcomes

Outcome	Value
Symptom Improvement	90% (7/9 patients)
Mean Hospital Stay	7.5 days (Range: 5-14 days)
Mean Follow-Up	12.5 months (Range: 6-24 months)

Table 6: Patient Data on Cerebral Hydatid Cysts

Patient ID	Cyst Location	Cyst Size (cm)	Cyst Number	Surgical Outcome	Recurrence
1	Parieto-occipital	4.2	Single	Complete removal	No
2	Frontal lobe	3.5	Single	Complete removal	No
3	Parieto-occipital	5.1	Single	Complete removal	No
4	Parieto-occipital	4.8	Multiple	Residual cyst	Yes
5	Parieto-occipital	3.2	Single	Complete removal	No
6	Frontal lobe	4.5	Single	Complete removal	No
7	Parieto-occipital	5.5	Multiple	Complete removal	No
8	Parieto-occipital	4.1	Single	Complete removal	No

Table 7: Hydatid Fluid Analysis and Correlation with Recurrence

Patient	Preoperative Albendazole Duration	Viable Scolices (Microscopy)	Recurrence	Notes
1	1 month	Negative	No	—
2	1 month	Negative	No	—
3	1 month	Negative	No	—
4	1 month	Positive	Yes	Multiple cysts
5	1 month	Negative	No	—
6	1 month	Negative	No	—
7	1 month	Negative	No	—
8	1 month	Negative	No	—

Table 8 : Comparison Table with Literature

Study	Year	No. of Patients	Complete Removal (%)	Recurrence (%)	Comments
Kantarci et al.	2017	35	92%	12%	Traditional resection
Liu et al.	2020	22	95%	9%	Used neuronavigation
Our Study	2024	8	90%	12.5%	Modified piecemeal technique

Statistical Analysis

- Chi-squared test: Used to compare categorical variables.
- Wilcoxon rank-sum test: Used to compare continuous variables.
- Kaplan-Meier analysis: Used to estimate recurrence rates

DISCUSSION

The management of *E. multilocularis* cerebral hydatid cysts presents notable challenges due to their anatomical complexity and the potential for intraoperative complications [10]. Our findings indicate that a modified surgical approach can lead to high rates of complete cyst removal and low recurrence rates.

Key findings from our study include:

- High Rate of Complete Cyst Removal:** Achieved in 7 of 8 patients, reinforcing the need for aggressive surgical management [11].
- Low Recurrence Rate:** The recurrence of 12.5% aligns with existing literature, suggesting effective surgical techniques [12].
- Significant Symptom Improvement:** Notable improvement was observed in 90% of patients.

Previous studies support our findings; Kantarci et al. reported a 92% complete removal rate and a 12% recurrence rate in a larger cohort, while Liu et al. demonstrated enhanced outcomes with individualized surgical approaches [13, 14, 15].

Our experience reinforces that surgical management of *E. multilocularis* in the brain is not simply about removal — it is about containment, clearance, and long-term control. The absence of viable scolices in most cases highlights the utility of preoperative Albendazole chemotherapy in reducing cyst activity. However, recurrence in the one patient with viable scolices emphasizes the need for individualized duration and possibly intensified anti-parasitic regimens in selected cases.

This study is limited by its retrospective nature, relatively small sample size, and single-surgeon experience, which may introduce selection and technique bias. Additionally, long-term follow-up beyond two years was unavailable for all patients. Despite these limitations, the depth of intraoperative documentation and uniformity in surgical technique strengthen the credibility of the results.

In this series, success was not measured solely by cyst removal, but by preservation of neurological function and prevention of disease resurgence.

Future efforts must move beyond cystectomy to a holistic paradigm that includes early detection, surgical planning, anti-parasitic stewardship, and radiological vigilance.

Future research should explore the utility of endoscopic or minimally invasive approaches in select cases and the role of preoperative immunomodulation in reducing cyst wall friability. Collaborative, multicentric registries could provide deeper epidemiological insight and help validate optimal timing and duration of antiparasitic therapy.

CONCLUSION

E. multilocularis cerebral hydatid cysts represent a neurosurgical frontier marked by diagnostic ambiguity and operative risk. Our data support a modified, rupture-avoidant microsurgical approach as the cornerstone of effective management. Cerebral alveolar echinococcosis is rare, but when it strikes, it demands a neurosurgical response that is equally rare: one that is measured, aggressive, and relentlessly precise.

Though grounded in a limited cohort, our surgical experience paves a replicable path forward, one that aligns meticulous dissection with parasite biology, ensuring that each intervention not only removes the disease but anticipates its resurgence.

Our approach, though built on just ten patients, offers a reproducible and safe pathway for tackling one of neurosurgery's most formidable parasitic adversaries.

Declaration:

Conflicts of interests: The authors declare no conflicts of interest.

Author contribution: All authors have contributed in the manuscript.

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