ISSN 1220-8841 (Print)
ISSN 2344-4959 (Online)

ROMANIAN
NEUROSURGERY

Vol. XXXX | No. 1 March 2026

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DOI: 10.33962/roneuro-2026-008

Romanian Neurosurgery (2026) XXXX (1): pp. 47-50
DOI: 10.33962/roneuro-2026-008
www.journals.lapub.co.uk/index.php/roneurosurgery

Successful management of double giant primary hydatid cyst of brain, that also revealed a cardiac location, in Yemeni centre: Case report

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Keywords
echinococcosis, hydatid cyst, intracranial hydatid cyst

ABSTRACT

Hydatid cyst is a parasitic infection caused by the larval stage of the *Echinococcus* tapeworm. It is endemic in various parts of the world, including the Middle East. The liver and lungs are the most commonly affected organs. Intracranial hydatid disease is rare, occurring in only 1–2% of all *Echinococcus granulosus* infections. We report a rare case of a hydatid cyst located in the brain, successfully treated with complete microsurgical excision in a 22-year-old male.

INTRODUCTION

Hydatid disease is a rare zoonotic infection caused by infestation with the larval stage of the cestode *Echinococcus granulosus*. Intracranial hydatid disease is uncommon, accounting for only 1–2% of all reported cases of hydatid disease [1]. The definitive hosts of *Echinococcus* are various carnivores, with dogs being the most common. Sheep, cattle, and humans serve as intermediate hosts. Humans typically become infected by ingesting the parasite's ova through contaminated fresh vegetables, drinking water, or direct contact with infected dogs [2,3]. The disease is endemic in several parts of the world, including the Middle East, Australia, New Zealand, South America, and Central and Southern Europe [4,5,6]. The liver is the most commonly affected organ (approximately 75% of cases), followed by the lungs (15%) and, less frequently, the spleen. Involvement of the central nervous system is extremely rare. When present, intracranial cysts usually involve the brain parenchyma, and in severe cases, may extend into the ventricular system [5]. Surgical excision remains the treatment of choice. Complete and intact removal of the cyst is essential to prevent recurrence and to avoid potential complications such as anaphylactic shock.

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ISSN 2344-4959 (online)
ISSN 1220-8841 (print)

First published
March 2026 by
London Academic Publishing
www.london-ap.uk

Case report

A 22-year-old male presented with complaints of headache and difficulty concentrating on studies for the past month. On physical examination, he was conscious without motor deficit but reported severe headache.

Neuroimaging

A computed tomography (CT) scan and magnetic resonance imaging (MRI) of the brain revealed two large cystic lesions in the right fronto-parietal region. The largest measured 7 × 4.4 × 6.5 cm and contained two peripheral hyperdense nodules. The lesions caused a mass effect with a 2.3 cm leftward midline shift and subfalcine herniation. There was no evidence of wall or nodular enhancement. The cysts appeared hypointense on T1-weighted images and hyperintense on T2-weighted images, while the nodules were isointense (figure 1).

Surgical Intervention

A right fronto-parietal craniotomy was performed, and the two cysts were successfully excised without rupture using Dowling's technique (figure 2). Postoperatively, the patient showed significant symptomatic improvement. Follow-up imaging confirmed complete resection of the cysts (figure 3).

Pathology and Additional Workup

Histopathological examination confirmed the diagnosis of hydatid cysts (figure 4). A chest X-ray and abdominal ultrasonography revealed no hydatid involvement in the lungs or liver.

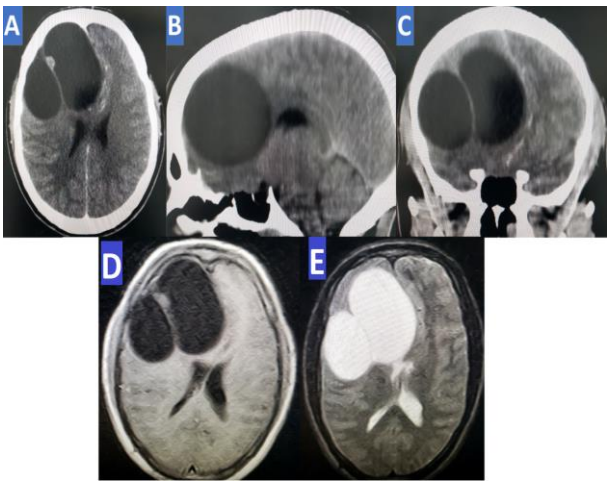

Figure 1. Pre-operative brain imaging: images. A: CT axiale, B: CT sagittal, C: CT coronal. D: MRI T1 WI. E: MRI T2 WI.

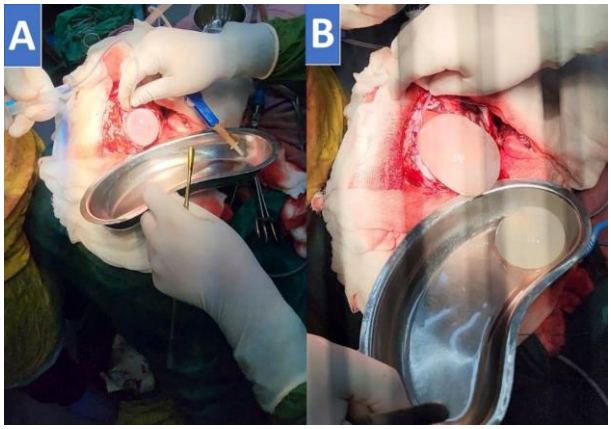

Figure 2. Peroperative images. A: extraction of the first cyst using hydro-dissection. B: extraction of the second cyst with the conservation of the cyst walls.

Figure 3. Post-operative CT: note the disappearance of the shift effect after cysts removal.

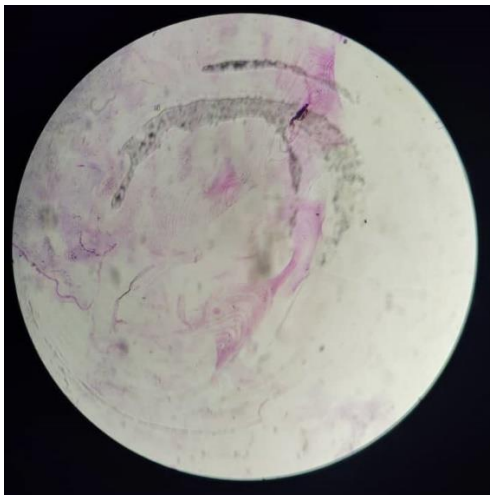

Figure 4. Pathology picture.

Postoperative Complications

Postoperatively, the patient was put under Albendazole for three months. He developed an intense, persistent cough unresponsive to medical treatment. Within a few days, cerebrospinal fluid (CSF) leakage occurred. The patient was reoperated three weeks later, and the dura was reinforced with galea tissue, resulting in complete cessation of the leakage. Due to the persistent cough, a cardiac ultrasound was performed, which revealed a cardiac hydatid cyst. The patient was referred to a specialized cardiac surgery unit. Unfortunately, he refused further treatment due to financial constraints.

Discussion

Intracranial hydatid cysts can be classified as primary or secondary. Primary cysts result from direct infestation of larvae in the brain, while secondary cysts are usually multiple and occur following the involvement of other organs. Although hydatid disease may be located anywhere in the brain, it is most frequently found in both hemispheres, particularly in the middle cerebral artery territory, with the parietal lobe being the most common site. Very rarely, the cysts are located in the posterior cranial fossa or ventricles [4,7]. In addition, intracranial hydatid disease represents 1–2% of all cerebral space-occupying lesions. Cerebral hydatid disease is usually diagnosed during childhood and is often solitary. Multiple cerebral hydatid cysts, though rare, usually result from dissemination secondary to spontaneous, traumatic, or surgical rupture of a primary cyst [4].

Lack of an effective immune system in the brain, special architecture of brain tissue, patent ductus arteriosus, and patent foramen ovale have been proposed as factors for isolated cerebral hydatid disease [1]. Radiology plays a key role in the diagnosis of hydatid cysts, although histopathological examination remains the gold standard [8]. Magnetic resonance (MR) and CT scans of the brain characteristically show a spherical, well-defined, and non-enhancing cystic lesion without peripheral edema. The fluid density is generally equal to that of cerebrospinal fluid on both CT and MR scans. Calcification of the wall is extremely rare, reported to be less than 1% of all cases. MR spectroscopy studies show pyruvate peaks along with lactate, alanine, and acetate [4,7]. The differential diagnosis should include pilocytic astrocytoma, which enhances on both CT and MRI and has a distinctive feature, which is the existence of a contrast-enhancing nodule. Arachnoid cysts are extra-axial in location and are not as round, considering there is no surrounding brain tissue. Abscesses have a thick wall, which vividly enhances with contrast, and they are surrounded by edema. In neurocysticercosis, multiple cysts are usually seen in different stages, and they have a pattern called “dot in a hole” since the larval scolex could be present as a mural nodule [2]. Surgical excision without rupture is the preferred treatment approach to prevent recurrence and anaphylactic reactions. The Dowling-Orlando technique is frequently employed for this purpose.

It requires a large osteoplastic flap, cautious handling during all the surgical steps, a wide incision over the cyst, sloping the head of the table downward, and injecting saline between the cyst and the brain [7,9]. Medical treatment with albendazole and praziquantel can be considered, particularly in recurrent cases or when rupture occurs during surgery [1,5,7]. Albendazole is administered as prophylactic chemotherapy after surgery, in a daily dose of 10 mg/kg three times a day for four months. Albendazole can also be administered prior to surgery to sterilize the cyst, minimize the risk of allergic reaction, and decrease the cyst wall's tension, lowering the likelihood of leakage during surgery and, as a result, the recurrence rate [10]. The most common complication is intraoperative cyst rupture, which may occur in 28% of cases, leading to dissemination into the subarachnoid space followed by severe inflammatory or anaphylactic response [9].

Conclusion

Primary brain hydatid cysts are uncommon; however, they must be considered as a differential diagnosis in endemic regions. Surgery is the treatment of choice and must be performed carefully to avoid spillage, followed by postoperative medication to reduce the risk of recurrence.

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