Radiology Case Reports 19 (2024) 4422–4425

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Hydatid cyst of the brainstem: The rarest of the rare locations for echinococcosis ☆

Ghassen Gader[a,*], *Abdelhafidh Slimane*[b], *Firas Sliti*[b], *Mansour Kherifech*[c], *Ala Belhaj*[b], *Asma Bouhoula*[b], *Jalel Kallel*[b]

[a] *University of Tunis-El Manar, Faculty of Medicine of Tunis. Department of Neurosurgery, Trauma and Burns Center, Ben Arous, Tunisia*

[b] *University of Tunis-El Manar, Faculty of Medicine of Tunis. Department of Neurosurgery, National Institute of Neurology, Tunis, Tunisia*

[c] *University of Tunis-El Manar, Faculty of Medicine of Tunis. Department of Surgery, Habib Bougatfa Hospital, Bizerte, Tunisia*

ARTICLE INFO

Article history:
Received 19 June 2024
Accepted 3 July 2024

Keywords:
Hydatid cysts
Brainstem
Neurosurgery
Radiology

ABSTRACT

Hydatidosis, caused by the larval stage of Echinococcus granulosus, is a zoonotic disease typically affecting the liver and lungs. Cerebral localizations are rare, especially in the brainstem. We present a case of a 9-year-old boy with a brainstem hydatid cyst. The patient exhibited progressive walking difficulties and limb impairment. MRI revealed a brainstem mass consistent with a hydatid cyst. Due to the lesion's size and location, surgical intervention was necessary. The cyst was decompressed and removed without complications, followed by albendazole treatment. Postoperative recovery was uneventful, and the patient showed no signs of recurrence after 2 years. This case highlights the importance of early diagnosis, precise surgical techniques, and thorough postoperative care in managing rare cerebral hydatid cysts.

Introduction

Hydatidosis is a zoonotic disease caused by the larval stage of Echinococcus granulosus (metacestodes). The disease is acquired by ingesting fresh vegetables or domestic water contaminated by the feces of definitive hosts, mainly dogs, containing eggs, or even after direct contact with these animals [1–4]. Although it is mainly encountered in the Mediterranean region, cases are still diagnosed worldwide [1,2]. Echinococcus is classified into 10 genotypes (G1-G10), but humans are mainly contaminated with Echinococcus granulosus (G1, G2, and G3) [1,5]. Hydatid cysts (HC) can be found in any organ of the body, with hepatic and pulmonary localizations being the most frequent [6]. While the liver and lungs are the most commonly affected, brainstem infection is exceedingly rare

☆ Competing Interests: The authors do not declare having any conflicts of interest regarding this manuscript.

* Corresponding author.
E-mail address: ghassgader@gmail.com (G. Gader).

https://doi.org/10.1016/j.radcr.2024.07.010

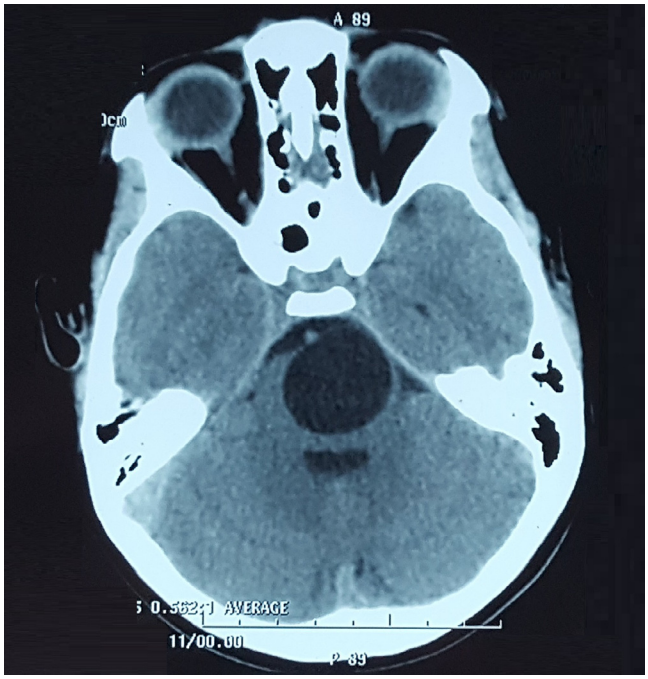

Fig. 1 – Axial section of a brain CT scan showing a round hypodense well circumscribed in the pons.

[7]. Cerebral localization of hyatidosis is uncommon, occurring in about 2% of cases, and it especially affects children [2,6]. These cysts are more frequently located in the supratentorial, mainly in the parietal lobe within the area supplied by the sylvian artery [1]. Hydatid cysts located in the brainstem are extremely rare. Here, we report a new case of a hydatid cyst located in the pons in a 9-year-old boy.

Case presentation

We report the case of a 9-year-old male patient with no significant medical history. The boy presented with a progressive onset of walking difficulties over the past 10 days, along with impairment of the left upper and lower limbs. He did not report any seizures, fever, or trauma.

On physical examination, the patient was conscious and exhibited right hemiparesis, as well as signs of right fifth (V) and seventh (VII) cranial nerve dysfunction. A brain CT scan (Fig. 1) revealed a hypodense lesion located within the pons, without associated upstream hydrocephalus. A subsequent brain MRI (Fig. 2) showed a brainstem mass measuring 31 × 26 × 27 mm, which was hypointense on T1-weighted imaging (WI) and hyperintense on T2-WI, without any enhancement after the injection of gadolinium. The lesion did not cause any surrounding edema and was protruding towards the left cerebellopontine angle. These features were consistent with a diagnosis of a brainstem hydatid cyst.

Due to the size of the lesion and the patient's clinical status, there were no alternatives to surgery despite the highly risked location of the lesion. Intraoperatively, a left subtemporal approach was performed to evacuate the contents of the lesion at its closest point to the pons surface. After puncture and aspiration, the cyst wall was progressively dissected from the surrounding neural structures.

The postoperative course was uneventful, and the patient regained full consciousness without any neurological sequelae. A follow-up CT scan (Fig. 3) confirmed complete removal of the cyst. Anthelmintic treatment with albendazole (12 mg/kg) was initiated, and the patient was discharged 4 days after surgery.

Two years postsurgery, the patient has shown no signs of recurrence and remains on albendazole.

Discussion

Cerebral localizations of hydatidosis are rare, representing 1%-4% of all hydatid cysts [8]. Most of these cysts are localized within the territory of the middle cerebral artery, especially the parietal lobes [7]. They are usually solitary, primary, and mainly diagnosed in children [1,7,9]. Posterior fossa locations are very rare [10,11]. Brainstem infections have only been reported in a few cases [2,7].

Clinical presentation of cerebral hydatidosis depends on the location of the cyst [4,6]. When located in the brainstem, the most common symptoms are nausea, headache, vomiting, weakness, visual disturbances, and signs of cranial nerve dysfunction [11,12]. Clinical evolution inevitably progresses towards worsening, and the cyst may quickly become life-threatening [2].

Radiological aspects of cerebral hydatid cysts are usually "typical" and leave few room for differential diagnoses [13]. On computed tomography (CT), a rounded, thin-walled, isodense and nonenhancing, lesion with no perilesional edema is usually found. Even if the interest of CT scan for posterior fossa lesions is relatively limited, brainstem hydatid cysts are usually highly suspected facing the features provided by this exam [6]. Brain MRI remains the gold standard for preoperative diagnosis of brainstem hydatid cysts. It shows a cystic lesion presenting the same signal intensity as the CSF: hypointense on T1-WI and hyperintense on T2WI, with no perilesional edema or contrast enhancement [2,14]. MRI also allows visualizing the capsule of the cyst: the pericyst [2,15]. MRI is also useful in identifying intracranial other parasitic cysts. In fact, cerebral hydatid cysts are generally solitary, but may be multiple in case of rupture that occurred following a trauma or a previous surgery [6,14].

Even if the radiological features for hydatid cysts are usually univoque, several clinical and radiological situations may lead to differential diagnosis issues. These differential diagnoses are mainly represented by arachnoid cyst, brain abscess, and pilocytic astocytomas [16,17]. Pilocytic astrocytoma contains nodules that enhance after contrast injection. Arachnoid cysts are not as round as hydatid cysts should be, and are extra-axial. Abscesses have a thick wall that enhances after contrast injection, and are surrounded by edema [2,18].

Surgery is the mainstay for treatment of cerebral hydatid cysts. The best technique is without contest is total "in block" removal avoiding peroperative spillage of cyst fluid, source for high risk of dissemination and anaphylactic shock [6,19].

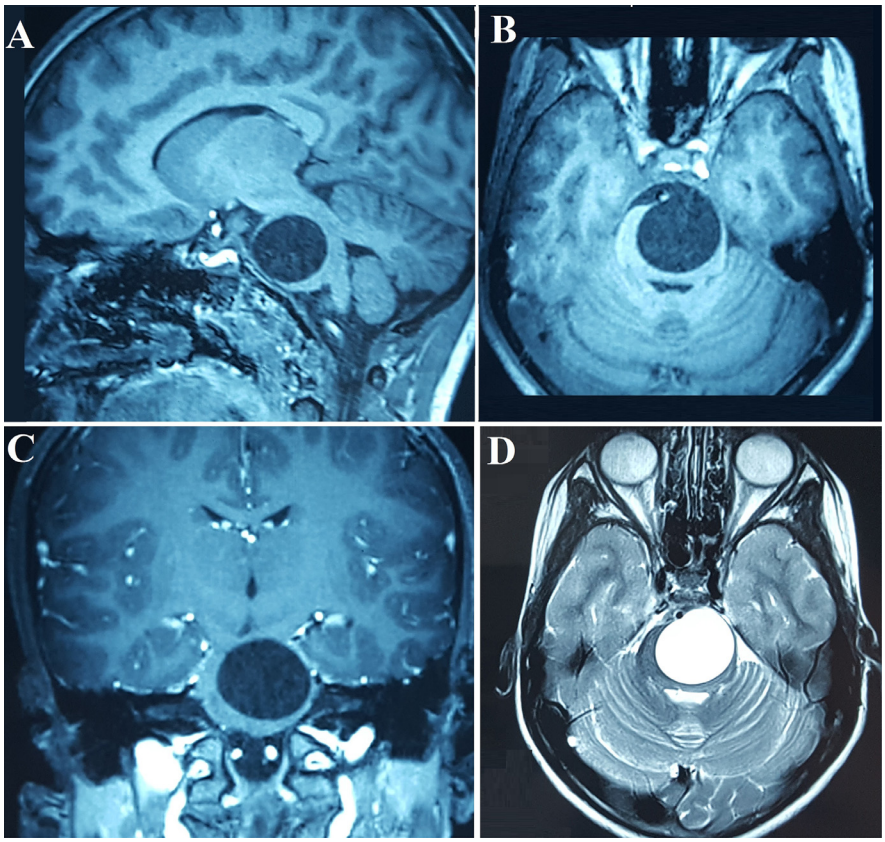

Fig. 2 – Sagittal (A), axial (B, D) and coronal (C) sections of a brain MRI on T2-WI (D) and T1-WI without contrast injection (A, B) and with contrast injection (C) showing a cystic lesion in the pons without any enhancement after injection of Gadolinium. The lesion slightly protrudes towards the left cerebellopontine angle.

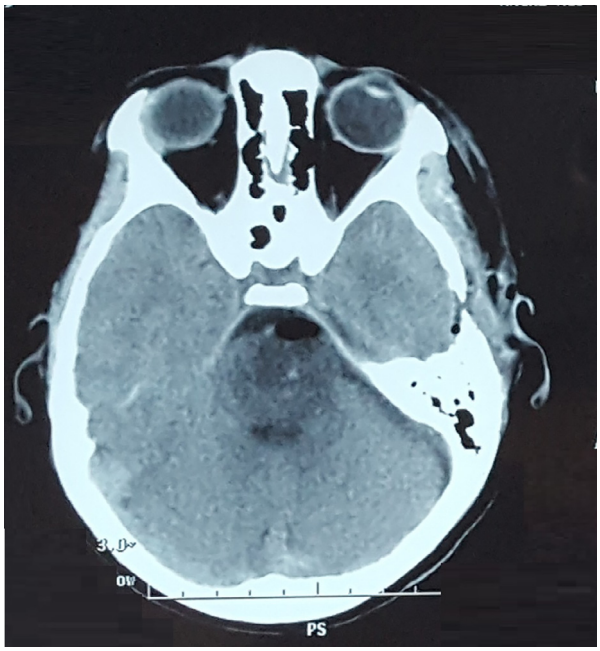

Fig. 3 – Axial section of postoperative brain CT scan showing a complete removal of the lesion.

Dowling's technique is the reference, based on a wide incision and craniotomy, and performing a hydraulic "delivery" of the cyst by gentle irrigation of the cyst-brain interface [20]. The use of valsalva technique and sloping the head of the table downward would help a safe evacuation of the cyst [2,21]. This approach is much more controversial for the cysts located in the brainstem. Some authors keep on using the same Dowling technique [6], which is evaluated to be source for high morbidity according to others [22,23]. In our experience, we preferred to decompress the cyst before dissecting and extirpating the membrane. This technique was also performed by other authors, who reported successful resection without anaphylaxis or dissemination [22,23]. However, in order to reduce the risk for recurrence after rupture, a generous wash of the surgical site should be performed using hypertonic saline and applying antihelminthics in situ [7,22].

Despite the benign nature of hydatidosis, critical localizations such as the brainstem may be source for significant mortality and morbidity. Postoperative mortality rate was estimated as 10% [2,7]. Antihelminthics such as ALBENDAZOLE are required in situations where rupture occurred during surgery, when patients present local recurrence or have extracerebral localizations witnessing the systemic character of the hydatidosis [1,7]. Some authors affirm the interest of maintaining patients under treatment long time after surgery in order to minimize the risk of recurrence and dissemination [16,21].

Conclusions

Hydatidosis is still endemic in certain regions, primarily affecting the liver and lungs. Cerebral localizations are rare, with posterior fossa and brainstem localizations being extremely uncommon. MRI is essential for accurate diagnosis and distinguishing these cysts from other conditions. Symptoms such as limb impairment and cranial nerve dysfunction indicate brainstem involvement and necessitate prompt intervention to prevent life-threatening progression. Surgery is the primary treatment, requiring meticulous techniques to avoid complications. Decompressing the cyst before removal can prevent issues like anaphylaxis or dissemination. Postoperative care with albendazole is crucial to prevent recurrence, and long-term follow-up is necessary. With appropriate surgical and postoperative care, patients can recover without neurological sequelae. This case underscores the importance of early diagnosis, careful surgical planning, and thorough postoperative management in treating challenging cerebral localizations of hydatidosis.

Patient consent

A signed consent has been obtained from the child's father regarding the publication of this article

REFERENCES

[1] Karakoç Z, Kasimcan M, Pipia A, Tore G, Alberti A, Varcasia A, et al. A life-threatening brainstem compression by cerebral Echinococcus granulosus. Infez Med 2016;24(1):62–6.
[2] Alok R, Mahmoud J. Successful surgical treatment of a brain stem hydatid cyst in a child. Case Rep Surg 2020;2020:1–3.
[3] Varcasia A, Tanda B, Giobbe M. Cystic echinococcosis in Sardinia: farmers' knowledge and dog infection in sheep farms. Vet Parasitol 2011;181:335–40.
[4] Deplazes P, Rinaldi L, Alvarez Rojas C. Global distribution of alveolar and cystic Echinococcosis. Adv Parasitol 2017;95:315–493.
[5] Nakao M, Lavikainen A, Yanagida T, Ito A. Phylogenetic systematics of the genus Echinococcus (Cestoda: Taeniidae). Int J Parasitol 2013;43(12–13):1017–29.
[6] Muthusubramanian V, Pande A, Vasudevan MC, Ravi R. Surgical management of brainstem hydatid cyst—an unusual site. Surg Neurol 2009;71(1):103–6.
[7] Turgut M. Intracranial hydatidosis in Turkey: its clinical presentation, diagnostic studies, surgical management, and outcome. A review of 276 cases. Neurosurg Rev 2001;24(4):200–8.
[8] Kehila M, Ammar N, Hattab C. Statistical study of hydatid locations. A propos of 644 cases–1980-1988. Tunis Médicale 1988;66(8–9):587–91.
[9] Ahmadi N, Badi F. Human hydatidosis in Tehran, Iran: a retrospective epidemiological study of surgical cases between 1999 and 2009 at two university medical centers. Trop Biomed 2011;28:450–6.
[10] Braham E, Bellil S, Bellil K. Hydatid cyst of the posterior fossa. Med Mal Infect 2007;37(5):281–3.
[11] Kayaoglu C. Giant hydatid cyst in the posterior fossa of a child: a case report. J Int Med Res 2008;36:198–202.
[12] Kizlica O, Altas M, Senol U, Oztek M. Hydatid disease located in the cerebellomedullary cistern. Case Rep Med 2014;2014:271365.
[13] Mascalchi M, Ragazzoni A, Dal Pozzo G. Pontine hydatid cyst in association with an acoustic neurinoma: MR appearance in an unusual case. Am J Neuroradiol 1990;12:78–9.
[14] Kohli A, Gupta R, Poptani H, Roy R. In vivo proton magnetic resonance spectroscopy in a case of intracranial hydatid cyst. Neurology 1995;45:562–4.
[15] Nurchi G, Floris F, Montaldo C, Mastio F. Multiple cerebral hydatid diseases: case report with magnetic resonance imaging study. Neurosurgery 1992;30:436–8.
[16] Polat P, Kantarci M, Alper F, Suma S, Koruyucu M, Okur A. Hydatid disease from head to toe. Radiographics 2003;23:475–94.
[17] Wani N, Kousar T, Gojwari T. Computed tomography findings in cerebral hydatid disease. Turk Neurosurg 2011;21:347–51.
[18] Şahin-Akyar G. Computed tomography and magnetic resonance imaging findings in cerebral hydatid disease. Radiography 2002;8(4):251–8.
[19] Ozek M. Complications of central nervous system hydatid disease. Pediatr Neurosurg 1994;20:84–91.
[20] Dowling E, Orlando R. Quiste hidatico del lobulo frontal derecho. Rev Esp Assoc Med Argent 1929;4:200.
[21] Carrea R, Dowling E, Guevara J. Surgical treatment of hydatid cysts of the central nervous system in the pediatric age (Dowling's technique). Childs Brain 1975;1(1):4–21.
[22] Boudawara M, Jemel H, Ghorbel M, Triki C, Soussi R. Hydatid cysts of the brain stem. Two cases. Neurochirurgie 1999;45(4):321–4.
[23] Donmez T, Bavbek M, Demiralp O, Arda N, Altinors M. Anterior pontine hydatid cyst: case report. Kobe J Med Sci 1998;44(2):45–50.