







Hydatid Cyst of the Cerebellopontine Cistern: Report of Two Cases with Literature Review

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Asian J Neurosurg 2023;18:800-804.

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Abstract

Keywords

- cerebellopontine cistern
- cerebral hydatid cysts
- ► Echinococcus granulosus
- magnetic resonance imaging
- retrosigmoid approach

Posterior fossa localization of a hydatid cyst is uncommon; in these cases, the cyst usually locates in the cerebellum. Localization within the subarachnoid spaces or the cerebrospinal fluid ventricular system is exceptional.

In the present report, which appears to be the seventh in the literature, we describe two cases of a hydatid cyst in the cerebellopontine cistern. Magnetic resonance imaging findings revealed a nonneoplastic cystic lesion mimicking an arachnoid cyst. The hydatid nature of the cyst was unexpected preoperatively. In both cases, the cyst was successfully removed using the puncture, aspiration, irrigation, and resection technique via a retrosigmoid approach. Histopathological examination confirmed the cysts to be Echinococcus granulosus in nature.

Hydatid cyst may be considered in the differential diagnosis of arachnoid cysts of the cerebellopontine cistern to determine which surgical procedure to perform and to avoid unexpected complications. Previous published cases were also discussed.

Introduction

Hydatid cysts of the posterior fossa are extremely rare. Moreover, most hydatid cysts are located intra-axially, that is, in the cerebellar hemisphere, fourth ventricle, or pons; those with an extra-axial localization, as in the cistern, are very uncommon.¹ Here, we present two cases of cerebellopontine (CP) cistern hydatid cysts with a special emphasis on magnetic resonance imaging (MRI) and the surgical procedure. A literature review of other six reported cases was also performed.

Case Presentation

Case 1

A 49-year-old man, previously healthy, presented with complaints about nausea, vomiting, headache, and gait distur-

article published online November 7, 2023

DOI https://doi.org/ 10.1055/s-0043-1775860. ISSN 2248-9614.

bance since a month before. The neurological examination revealed left-sided sixth and seventh nerve palsies and left cerebellar signs. MRI showed an extra-axial cystic lesion in the left CP cistern that was hypointense on T1-weighted sequences and hyperintense on T2-weighted sequences with enhancing cyst wall. In addition, the cyst ($32 \times 20 \times 44 \text{ mm}$) compressed the brain stem and the fourth ventricle. MRI also confirmed pericystic edema better seen on fluid-attenuated inversion recovery (FLAIR) sequences (Fig. 1). MRI suggested the nonneoplastic nature of the lesion, as in an arachnoid cyst. The lesion was exposed through the retrosigmoid approach for marsupialization of what we thought was an arachnoid cyst. After opening the arachnoid membrane, another distended dense cystic membrane was seen. At this time, a hydatid cyst was suspected of glistening white aspect of the cyst and because we are in endemic geographic

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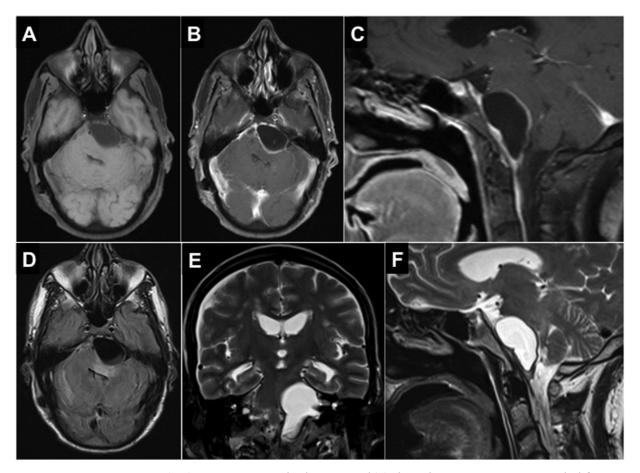


Fig. 1 Magnetic resonance imaging (MRI) Precontrast T1-weighted images, axial (A), show a hypointense cyst occupying the left cerebellopontine (CP) cistern. The cyst has considerable mass effect on brain stem and the fourth ventricle is collapsed. Postcontrast T1-weighted image (B and C) shows contrast enhancement of the cyst wall, possibly related to the presence of vital scoleces and to the inflammatory pericystic reaction. Fluid-attenuated inversion recovery image (D) shows hypointense cyst with circumscribed hyperintense area, probably representing edematous or inflammatory changes, is seen posterior to the cyst. MRI T2-weighted image, coronal (E) and sagittal view (F), shows signal within left CP cistern cyst to be hyperintense compared with cerebrospinal fluid.

areas of this parasitosis. Therefore, the water jet dissection technique as surgical treatment is not useful in the CP angle due to the adhesion of the cyst to several important nervous structures. A fine needle was inserted into the cyst under meticulous caution not to rupture the cyst to prevent dissemination and the content was aspirated gently until the cyst was totally decompressed. The cyst wall remained intact and was totally dissected from the adjacent structures microsurgically. The cyst cavity and surrounding neural tissue were irrigated with a normal saline solution for a few minutes, and the cyst wall was totally removed. Histopathological examination confirmed the diagnosis of hydatid cyst (>Fig. 2). Albendazole was given postoperatively at the dosage of 15 mg/kg per day and continued for 3 months. Cranial nerves abnormalities improved markedly in this period.

Case 2

A 39-year-old man was admitted to our department with a 3-month history of progressive inability to walk, headache, and dizziness. Neurologic examination revealed right cerebellar signs, ataxia, and nystagmus. MRI demonstrated a cystic lesion $(42 \times 32 \times 45 \text{mm})$ in the right CP cistern that

exerted a pressure effect on the brain stem and the fourth ventricle without hydrocephalus. The lesion was hypointense on T1-weighted and FLAIR images, hyperintense on T2-weighted images with the same signal as cerebrospinal fluid (CSF), which is suggestive of an arachnoid cyst. The lesion had no enhancement after gadolinium administration. MRI also confirmed pericystic edema better seen on T2-weighted and FLAIR sequences (Fig. 3). Cystic lesion was removed with the microsurgical technique at right CP angle using retrosigmoid approach. After opening the arachnoid membrane at the lateral CP cistern, a cystic lesion was found, firmly attached to cranial nerves and arachnoid membranes. At this time, a hydatid cyst was suspected. Therefore, the cyst was punctured, and the fluid was aspirated with a needle to prevent contamination. The cyst cavity and surrounding neural tissue were irrigated with a hypertonic saline solution for a few minutes, and the cyst wall was totally removed. Histopathological examination confirmed the cysts to be hydatid in nature. Postoperative course was uneventful; he was discharged from the hospital on postoperative day 8 and was also treated with albendazole (15 mg/kg per day) for 3 months with regular follow-up.

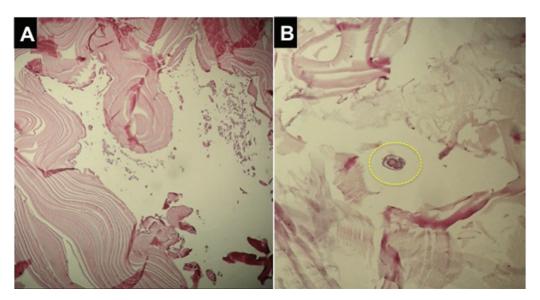


Fig. 2 (A) Microscopic image of laminated membrane of hydatid cyst (hematoxylin and eosin stain, x60). (B) Microscopic image of a scolex (dotted circle) surrounded by laminated membrane lining the hydatid cyst (hematoxylin and eosin stain, x60).

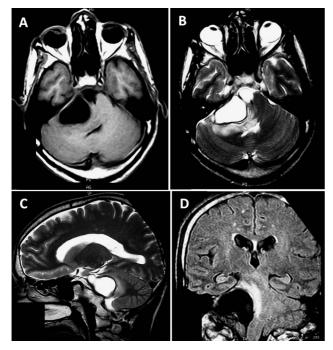


Fig. 3 Magnetic resonance imaging (MRI) demonstrated a cystic lesion in the right cerebellopontine cistern that exerted a pressure effect on the brain stem and the fourth ventricle without hydrocephalus. The lesion was cystic and hypointense on T1-weighted images (A), hyperintense on T2-weighted images with the same signal as cerebrospinal fluid, which is suggestive of an arachnoid cyst (B and C). The lesion had no enhancement after gadolinium administration. MRI also confirmed pericystic edema better seen on T2-weighted and fluid-attenuated inversion recovery sequences (B–D).

Discussion

Central nervous system involvement in the hydatid cyst disease is found in approximately 1 to 2% of all the cases of hydatid disease, and 50 to 75% cases of intracranial hydatid cyst are seen in children.² Hydatid cysts of the posterior fossa are extremely rare. Moreover, most hydatid cysts are located

intra-axially, that is, in the cerebellar hemisphere, fourth ventricle, or pons; those with an extra-axial localization, as in the CP, are very uncommon. Only six cases were previously reported. The blood supply of the CP angle is limited and thus the incidence of the hydatid cyst in this specific location is extremely low.

MRI is the best neuroimaging choice to diagnose a hydatid cyst. The cystic signal is iso or hypointense on T1-weighted and hyperintense on T2-weighted images. The lesion typically shows no contrast enhancement, and calcifications, usually peripheral, are rare. However, MRI may also show pericyst as a halo of high signal intensity on T2-weighted images, and, in some cases, perifocal edema may be detected. In one case, we observed a faint ring enhancement within the cyst, in both cases, there was high signal pericyst intensity on T2-weighted images as a perifocal edema. These unusual features could be attributable to pericystic inflammatory reaction and led us a posteriori to expect the preoperative diagnosis of hydatid cyst.

Regarding the differential diagnosis of hydatid cysts, arachnoid cysts and epidermoids are common in the CP cisterns. Cystic astrocytomas, porencephalic cysts, and pyogenic or fungal abscesses are other lesions to be differentiated. Epidermoid cysts usually have slightly hyperintense signal intensity on proton density MRI. These lesions usually surround neighboring nerves and vessels, whereas arachnoid and hydatid cysts displace adjacent structures. Arachnoid cysts and porencephalic cysts do not show contrast enhancement as much as hydatid cysts. However, hydatid cysts can have wall enhancement. On the other hand, diffusion MRI is not very useful because both hydatid and arachnoid cysts show increased diffusion. ¹⁰

The ideal treatment for cerebral hydatid cysts is surgical, and the cyst must be removed unruptured using water jet dissection technique (Dowling technique), or "hydatid birth"; this technique is applicable in almost all cerebral hydatid cysts according to the fact that the cyst does not have

Table 1 Summary of clinical characteristics of eight cases with hydatid cyst of the CP cistern reported in English literature up to date including our cases

Author (reference)	Year of publication	Country	Age (year)/sex	Multiplicity of the cyst	Location	Clinical presentation	Other organs location	Treatment	Outcome/ follow-up
Akdemir et al ³	2007	Turkey	19/M	Multiple	Right CP cistern, internal acoustic canal and jugular foramen	ICP, right V–VII, XI nerve palsies, and cerebellar signs	Liver	Surgery	Good without recurrence/ 3 months
Kizilca et al ⁶	2014	Turkey	45/F	Solitary	Left cerebellomedullary cistern	ICP, monoparesis in the left lower extremity	None	Surgery	poog
Karakoç et al ⁵	2016	Turkey	14/F	Multiple	Bilateral CP cistern	Respiratory distress, progressive quadriparesis	None	Surgery + Albendazole	Good without recurrence/ 1 year
Taghipour et al ⁷	2017	Iran	62/F	Solitary	Left CP cistern, Meckel's cave	Left V, VI, VII nerves palsies, hearing loss	None	Surgery + Albendazole	Good without recurrence/ 2 years
Alkhotani et al ⁴	2019	Saudi Arabia	44/F	Multiple	Left CP cistern, foramen magnum	Headache, left V–IX, XI, and XII nerves palsies, cerebellar signs	None	Surgery + Albendazole	Good without recurrence/ 1 year
Dere et al ⁸	2022	Turkey	68/F	Solitary	Left CP cistern	Difficulty in swallowing, negative gag reflex, and left cerebellar signs	None	Surgery + Albendazole	Lost to follow-up
Case 1	I	Могоссо	49/M	Solitary	Left CP cistern	ICP, left VI, VII nerve palsies, and left cerebellar signs	None	Surgery + Albendazole	Good without recurrence/ 3 months
Case 2	ı	Могоссо	39/M	Solitary	Right CP cistern	Headache, nystagmus, and right cerebellar signs	None	Surgery + Albendazole	Good without recurrence/ 2 years

Abbreviations: CP, cerebellopontine; ICP, intracranial pressure.

adhesions to the surrounding pia matter.¹¹ However, this method is not useful in CP angle due to adhesion of the cyst to several neuronal structures (cranial nerves, brain stem, and the cerebellum blood vessels).

Following a careful review of the published articles of the hydatid cyst of the CP cistern up to date, using PubMed search engine we found only six cases.^{3–8} The clinical characteristics of those cases (demographics, clinical presentation, localization, treatment plan and outcome) were analyzed and gathered in a summarized ► Table 1. Anatomically, all of the cases occurred in CP cistern involving in some cases bilateral CP angle, internal acoustic canal, jugular foramen, Meckel's cave, and foramen magnum. The cyst was solitary in three cases and multiple in three cases; also an enhancement of the cyst wall was seen in only one case. In both of our cases, the cyst was solitary. Treatment plan is almost always the same with careful surgical resection using the microsurgical technique through the retrosigmoid approach. Novel technique was reported in one case consisting of needle aspiration and internal decompression along with dissection of the cyst microsurgically without rupture it. This novel technique was successful and there was no residue. Most cases are treated with Albendazole for at least 6 months. The prognosis after surgical removal was excellent in all of the cases reported without recurrence.

Conclusion

We have presented rare cases of a hydatid cyst in the CP cistern (unsuspected preoperatively). Hydatid cyst should be considered in the differential diagnosis of cystic lesions of the CP cistern. Despite the benign nature of hydatid cyst, invasion of critical areas such as CP cistern may cause serious complications. MRI clearly demonstrated cisternal, neural, and vascular relationships that aided in intact surgical removal of the lesion using microsurgical techniques. Total removal without rupture should be the surgical goal in all hydatid cysts. Prognosis is generally excellent.

Data Availability Statement

All data generated and analyzed in this study are included in this article.

Consent for Publication

Both the patients provided written informed consent for publication of patient clinical details and clinical images.

Authors' Contributions

H.B. contributed to the conception, drafting, and reporting of the cases. S.B. acquired the clinical data. M.B. and A.A. contributed to the revision of the manuscript. All authors have read and approved the final manuscript.

Conflict of Interest None declared.

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