Anterior Interhemispheric Transcallosal Transchoroidal Fissure Approach for Third Ventricle Hydatid Cyst: An Interesting Approach to a Rare Entity

Seyed Ali Mousavinejad1, Hamid Borghesi-Razavi1, Ahmad Jabbari1, Mohammad Samadian3, Kristen Almagro2*, PAParviz Karimi1 and Omidvar Rezaei5

1Skull Base Research Center, Loghman Hakim Medical Center, Shahid Beheshti University of Medical Sciences, Iran
2Department of Neurosurgery, Pauline Braathen Neurological Center, Cleveland Clinic Florida, USA
3Department of Neurosurgery, Loghman Hospital, Shahid Beheshti University of Medical Sciences, Iran
4Department of Pediatric Neurology, Ilam University of Medical Science, Iran
5Department of Neurosurgery, Skull Base Research Center, Loghman Hakim medical center, Shahid Beheshti University of Medical Sciences, Iran

Abstract

In this case report, we describe the first case of transcallosal removal of a rare third ventricle hydatid cyst, which can be considered in the differential diagnosis of suprasellar arachnoid cysts. Caution must be taken in determining the best surgical approach to prevent unexpected complications. A primary intraventricular hydatid cyst is a rare phenomenon, which is most commonly in the lateral ventricle. After thorough research, only two cases of hydatid cyst have been discovered in the third ventricle. Therefore, we present an interesting case of a three-year-old girl with nausea, vomiting, and progressive drowsiness that upon neuroradiological examination was found to have a large, spherical, well-defined cystic lesion within the third ventricle. The patient underwent surgery via anterior interhemispheric transcallosal approach, and the cyst was successfully removed using Dowling’s technique.

Introduction

A hydatid cyst is a chronic Zoonotic infection, which are produced by larval cestodes of Echinococcus granulosus. Humans can become infected as intermediate hosts of these tapeworms through accidental ingestion of Echinococcus eggs [1-3].

Involvement of the Central Nervous System (CNS) as the primary location of human echinococcosis is a rare phenomenon, accounting for 2% to 4% of all echinococcosis infections [4,5]. Among primary intracranial hydatid cysts, a cerebral intraventricular hydatid cyst is very rare. To the best of our knowledge, only 37 cases of intraventricular hydatid cysts have been reported, most commonly in the lateral ventricle. Even more rarely, only two cases of third ventricle involvement have been reported. Herein, we report the third case of a primary single hydatid cyst located in the third ventricle in a child presenting with hydrocephalus. The cyst was resected using the anterior interhemispheric transcallosal approach.

Case Presentation

A three-year-old girl presented to the emergency department with nausea, vomiting, drowsiness for two days. She reported a five-month history of headache, which had intensified significantly during the one week prior to admission. Upon admission, the patient was increasingly difficult to arouse, however the brainstem and spinal reflexes were intact, and there were no lateralizing neurological signs. Fundoscopic exam revealed bilateral Papilledema. Hematological studies performed at an outside hospital were within the normal limit. There was no family history of hydatid disease. Brain Computed Tomography (CT) scan (Figure 1a) and Magnetic Resonance Imaging (MRI), conducted in the previous hospital, revealed enlarged lateral and third ventricles as well as a round cystic lesion (35 mm × 40 mm) in the third ventricle. Brain MRI also showed a large, spherical, well-defined, cystic lesion within the third ventricle. The signal intensity of the cyst content was similar to that of Cerebrospinal Fluid (CSF) (hypointense in T1-weighted MR and FLAIR, hyper intense in T2-
A cyst was removed intact, using Dowling’s technique of gravitational position, and the patient’s head was turned towards the floor. The operating table was placed in the Trendelenburg position between the cyst wall and the lateral wall of the third ventricle laterally. A transchoroidal dissection was performed to expose the superior wall of the ventricle and obstructed the foramina of Monro. A Foley catheter was placed gently between the cyst wall and the lateral wall of the third ventricle (Figure 3b).

The cyst was removed intact, using gravitational forces and hydrostatic expulsion (Dowling’s technique) (Figure 4a, 4b). The ventricular cavity was irrigated immediately with Ringer’s lactate solution. The patient recovered without complications, and post-operative CT scan confirmed complete removal of the lesion. After surgery, the patient underwent systemic work-up of other organs. The results of abdominal and pelvic ultrasonographic and echocardiographic studies were normal. The result of indirect hem agglutination test was also negative for hydatid disease. In the 10th month follow-up after surgery, the results of neurological examination were within the normal limits.

Discussion

In hydatid disease, which is recognized as a Zoonotic disease, humans are an intermediate/accidental host, while dogs are definitive hosts. In this condition, humans become infected by ingestion of food, milk, or water, contaminated with dog feces, containing the ova of parasite. In humans, it is commonly caused by Echinococcus granulosus and less frequently by Echinococcus multilocularis [1,2,4]. It is endemic to the Middle East, Mediterranean countries, South America, North Africa, and Australia. Therefore, Iran, as a Middle Eastern country, is an important endemic focus of hydatid disease [6,7]. Brain involvement is rare in hydatid disease, accounting for about 2% to 4% of all human hydatid diseases, even in endemic areas [5]. This condition is more common in children, as more than 70% of intracranial hydatid cysts occur in this population [8,9]. Intracranial hydatid cysts may be categorized into primary and secondary types. Primary hydatid disease is the most common type, which usually manifests as a simple lesion. In the primary type, brain involvement is caused by direct infestation of larvae in the brain, without demonstrable involvement of other organs, while the secondary type usually involves multiple lesions and is a result of spontaneous, traumatic, or surgical rupture of the primary intracranial hydatid cyst or embolization of a ruptured cardiac cyst [10,11]. Intracranial hydatid cysts are slowly growing lesions at a rapid rate.
of 1.5 to 10 cm per year [12,13]. Because of their low growth rate, they usually appear late when they are large enough to cause mass effect. Clinical presentation depends on the size and location of the lesion. Headache and vomiting are the most common presenting signs, while neurological deficits can occur due to increased intracranial pressure and epileptic seizure in the late stage [5,14]. Cerebral hydatid cyst is generally located in the cerebellar hemispheres (75%) in the middle cerebral artery territory [15]. However, few documented cases of unusually located intracranial hydatid cysts in the thalamus [16,17], parasellar region [18], interpeduncular cistern [5] cavernous sinus [19], and brainstem [20,21] have been reported. Ventricular involvement is unusual in brain hydatid cysts. The lateral ventricle is the most common site, while the third ventricle involvement is extremely rare. There is no consensus about the exact mechanism of ventricular involvement in these patients, but it seems that ionsopores reach the ventricular cavity through the choroid plexus [22,23]. Other researchers believe that spillage of ruptured primary cysts near the ependymal lining into the ventricle causes multiple secondary cysts [24]. In nearly 20% of patients with hydatid disease, other lesions may be found in other organs [25]. A complete radiological evaluation after surgery, including abdominal and pelvic ultrasonography, echocardiography, and chest X-ray, should be performed postoperatively. In our case study there was no evidence of systemic disease. In patients with cerebral hydatid disease, Weinberg and Casoni serological tests have little practical values in confirming the diagnosis. The results of both tests were negative in our patient [14]. Detection of anti- *Echinococcus* IgG antibodies, using Enzyme-Linked Immunosorbent Assay (ELISA), is the definitive laboratory diagnosis of hydatid disease. However, in isolated cerebral hydatid cysts, specificity and sensitivity of this test are reduced by nearly 30%. In our case, the findings of ELISA assay were negative [2,26]. In CT scans, the cysts present as hypodense, rounded mass lesions, without perifocal edema. MRI can reveal well-defined, smooth, thin-walled, spherical, homogeneous cystic lesions. Cystic fluid is similar intensity to CSF on T1- and T2-weighted images. The cyst wall usually shows a rim of low-signal intensity on both T1- and T2-weighted images. Rim enhancement, perifocal edema, and calcification are rare features of hydatid disease [27-29]. Magnetic Resonance Spectroscopy (MRS) has been employed to identify the progression of hydatid cyst, which is associated with an increase in the amount of succinate, lactate, alanine, and acetate, a large pyruvate resonance, and an increase in myo-inositol level in these patients. However, these results are not confirmatory considering the specificity and sensitivity of 70% [30]. Based on radiological findings, differential diagnoses of cerebral hydatid cyst in the third ventricle include ependymal cyst, arachnoid cyst, and colloid cyst. Although most colloid cysts are solid, cystic lesions have been reported in few cases. Lack of ring enhancement, mural nodules, and perifocal edema excludes abscess and cystic astrocytoma. In contrast to hydatid cysts, suprasellar arachnoid cysts are not spherical. They displace the chiasm and infundibulum anteriorly [26,31]. Since the cyst in our case was round and located solely in the third ventricle, preoperative diagnosis of a third ventricle hydatid cyst was made. To the best of our knowledge, this is the third case of third ventricle hydatid cyst in a patient with hydrocephalus. The first case was reported by Prasad et al. in a 20-year-old man with a three-month history of headache, who underwent cyst resection by endoscopy [32]. The second case was reported by Habibi et al. [33] in a 16-year-old female with a three-year history of seizures, who underwent Venticuloperitoneal shunting. In the follow-up, her condition progressed to intractable generalized tonic-clonic seizure, and her level of consciousness reduced two years after shunt placement. She underwent surgical removal of the cyst, using a posterior parietal cortical incision towards the atrium of the left lateral ventricle. In the present case, considering the cyst location, we performed surgery using the anterior interhemispheric transcallosal approach. Two important points in this approach which can result in complications include drainage of the cortical vein, especially vein of Trolard, to the superior sagittal sinus (limiting our surgical field) and location of the internal cerebral vein relative to the cyst. Considering all aspects of the cyst location, we decided to perform large front parietal craniotomy using the transcallosal approach with caution. If during surgery, the cortical vein is in our field of view, it is necessary to change the approach to the frontal transcallosal approach with PAIR technique. If during dissection of the choroidal fissure, the internal cerebral vein is in the superior wall of the cyst (preventing its displacement for tumor removal); needle aspiration and irrigation must be employed. However, during surgery, according to preoperative MRI the draining cortical vein was placed 1.5 cm behind the coronal suture in our case. We used a wide field for collosal dissection, and we could easily see the upper anterior and posterior aspects of the cyst wall for wide choroidal dissection. The cyst pushed the internal cerebral vein to the posterior aspect. Therefore, we could place a Foley catheter between the cyst wall and the lateral wall of the third ventricle and apply the Dowling’s technique (Figure 3). Surgery has been the best therapeutic approach for the removal of brain hydatid cyst [34]. Considering the location of the cyst and the patient’s medical condition, there are multiple surgical options available. In general, for lesions located in the superficial area, which are not in the eloquent region, the Dowling’s technique, i.e., extraction by forcing saline solution around the lesion (hydrostatic expulsion), is the most accepted choice [35].

The PAIR technique involves puncture with a needle, followed by aspiration, irrigation, and resection, reserved for the problematic lesion, such as brainstem lesion. However, the surgical outcomes of PAIR technique are not as good as Dowling’s technique [20,36]. The most important point in surgery is prevention of cyst rupture, which can occur in 16.9% to 25.6% of cases, and is more common in deep-seated lesions. It can lead to anaphylactic shock and meningitis. If rupture occurs in the ventricular system, multiple hydatid cysts along the CSF system may occur [37-39]. Generally, medical treatment is not an alternative option for surgery. Nevertheless, medical treatment with albendazole can be used for patients with multiple cerebral lesions or those with poor conditions, who are not proper candidates for surgery. It is also useful for cases of intraoperative rupture of cyst and recurrence of hydatic disease.

Some authors believe that pre-surgical administration of albendazole for a short period before surgery can reduce the risk of anaphylactic reaction and recurrence [40,41]. In our case, given the patient’s emergency condition, after consulting with an infectious diseases specialist, we decided not to administer albendazole before surgery.

**Conclusion**

Intracranial hydatid cyst is an unusual form of hydatid disease, which can be seen in all parts of the brain, including the ventricular system. Given its low growth rate, clinical presentations may appear late when the cyst has a large size, therefore, the risk of surgical complications may increase. Despite their rarity, with precise
evaluation of neuroimaging findings, we can differentiate hydatid cysts from other cystic pathologies to prevent mismanagement and surgical complications.

References


