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# Multiple Intracranial Hydatid Cysts in a Boy

## Bir Erkek Çocuğunda Birden Fazla İntrakranyal Hidatik Kist

Halit ÇAVUŞOĞLU
Cengiz TUNCER
Ahmet ÖZDİLMAÇ
Yunus AYDIN

Şişli Etfal Training and Research Hospital, Neurosurgery Clinic, İstanbul, Turkey

**ABSTRACT**

Multiple hydatid cysts of the brain are uncommon and may be either primary or secondary. A 15-year-old boy with a huge mass of intracranial hydatid cysts (95x85x80 mm) is presented. The first manifestation was headache and vomiting, which was followed by symptoms of raised intracranial pressure. The patient underwent an urgent operation due to rapidly deteriorating neurological status, and 19 hydatid cysts were removed. Unfortunately, the patient's neurological status did not improve and he died. Hydatid cyst is a benign lesion. Surgery is the standard and most effective treatment for intracranial hydatid cysts. Appropriate and timely management is mandatory for reducing the mortality and morbidity.

**KEYWORDS:** Brain, Hydatid cyst, Multiple cysts

**ÖZ**

Beynin multipl hidatik kistleri seyrek olup primer veya sekonder olabilir. 15 yaşında erkek çocuğunda devasa yer kaplayıcı intrakranyal hidatik kistler (95x85x80 mm) sunulmaktadır. Başağrısı ve kusma ilk olarak ortaya çıkmış ve kafa içi basınç artışı bulguları bunları takip etmiştir. Olgu nörolojik durumundaki hızlı kötüleşme nedeniyle acilen ameliyata alındı ve 19 adet hidatik kist çıkarıldı. Maalesef hastanın nörolojik durumunda düzelme olmadı ve hasta öldü. Hidatik kist selim karakterli bir lezyondur. Mortalite ve morbiditeyi azaltmak için uygun ve zamanında müdahale şarttır.

**ANAHTAR SÖZCÜKLER:** Beyin, Hidatik kist, Multipl kist

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Correspondence address:
**Halit ÇAVUŞOĞLU**
E-mail: anhcavusoglu@yahoo.com

## INTRODUCTION

Cerebral hydatid disease is rare and occurs in about 2% of cases (35). Hydatid cysts of the brain are usually single, spherical, unilocular, and may be large; in rare instances, they can be multiple and embolic (34). Although an intracranial single lesion is nearly always primary, multiple lesions are frequently secondary (4,5,12,16,19). Multiple hydatid cysts resulting from the rupture of a primary cyst are acephaloceles; they are infertile and have no broad capsule. However, very rarely a multiple larval intake may cause primary multiple cerebral hydatid cysts (3,4,10,19). Intracranial hypertension secondary to mass effect is usually the first clinical sign of brain involvement. Because of their indolent nature, hydatid cysts may not cause focal neurological signs until they are very large (35). They often become significantly larger, particularly in children. In this report, we describe a huge hydatid cyst mass, whose large size had apparently raised the intracranial pressure, causing death.

## CASE REPORT

A 15-year-old boy from a rural area was admitted to our neurosurgery department in a state of coma. Neurological examination revealed absence of the pupillary reflex in the right eye. Glasgow Coma Scale (GCS) score was 5 (E=1, M=3, V=1). The patient had been admitted to a hospital in another city with complaints of headache and vomiting four weeks previously. Magnetic resonance imaging (MRI) demonstrated a huge (95x85x80 mm) mass of multiple cystic space-occupying lesions in the right parieto-occipital region, with midline shift to the left hemisphere (Figure 1). The community doctor referred the boy to a neurosurgery clinic with these symptoms.

He was admitted to our emergency service in a state of coma. Patient underwent axial cranial computed tomography (CT) after initial airway and hemodynamic management. CT scan showed multiple hydatid cysts occupying the right parieto-occipital region. No peripheral edema was noted around the cysts (Figure 1). The patient was immediately operated on. A large craniotomy was performed. A soft rubber catheter was inserted between the hydatid cyst and surrounding brain tissue, and warm saline was injected through this catheter [Dowling's technique (7)]; 19 cysts were removed unruptured. Multiple hydatid cyst vesicles ranging from 20 mm to 60 mm in diameter were present (Figure 2). The pseudocyst capsule was then completely removed.

An early postoperative follow-up CT scan revealed cyst-free intracranial content (Figure 3). The thorax and abdomen were also observed with CT scans. No lesion was seen on careful examination of the liver, lungs, and other organs. Serological tests for hydatid disease were negative. Eosinophil count was within normal limits. Albendazole was

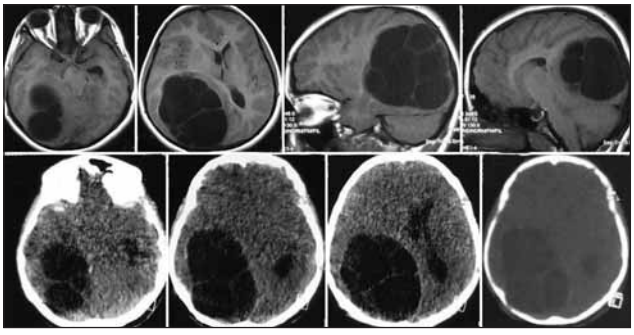

***Figure 1:*** *MRI of the brain (upper), showing a large mass of multiple cystic space-occupying lesions in the right parieto-occipital region, with midline shift to the left hemisphere. Edema is not seen in the surrounding brain tissue. Cerebral axial CT scan (lower), showing multiple hydatid cysts occupying the right parieto-occipital region with a thinning of occipital bone.*

***Figure 2:*** *Hydatid cysts and pseudocyst capsule removed at operation.*

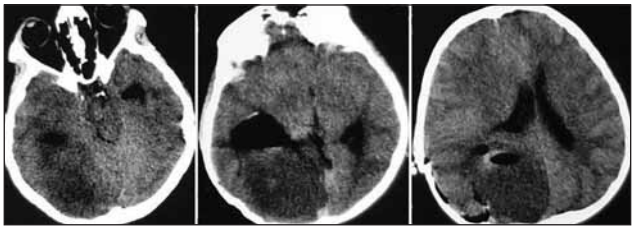

***Figure 3:*** *Cystic masses are not seen on early postoperative control CT scan.*

administered to the patient 10 mg/kg twice daily. Unfortunately, the patient's neurological status did not improve and he died on the sixth postoperative day.

## DISCUSSION

Cerebral cystic echinococcosis is seen in approximately 2% of patients infected with the parasite and this represents only 2% of all intracranial space-occupying lesions. It is most commonly seen in children and young adults (approximately 50–70%) (9,35).

Multiple hydatid cysts of the brain are very rare. Primary multiple cysts of the brain resulting from arterial embolism secondary to ingestion of multiple larvae and without any radiological or clinical evidence of hydatid disease elsewhere in the body are extremely rare (3,4,10,13,18,19,20,25,26,29,30,31, 32,33,34,37,39). In a review of the literature related to primary multiple intracranial cerebral hydatid cysts, we were able to find only 17 cases reported up to the time of preparing this article (Table I). Secondary multiple hydatid cysts of the brain can result from spontaneous, traumatic, or surgical rupture of a primary solitary cerebral cyst or as a consequence of a cyst rupture elsewhere and embolization of hydatids to the brain (14,15,21,38). Multiple hydatid cysts resulting from the rupture of a primary cyst are acephaloceles; they are infertile and have no broad capsule and scolices (2,4,5). The growth rate of cerebral hydatid cysts is 1 cm in diameter per year. This rate, however, may be more rapid in children (1,4,9,16,18,35).

Headache and vomiting and, motor weakness due to increased intracranial pressure are the most common clinical manifestations of hydatid disease (22,28). Headache is usually the earliest symptom. The serologic tests are of little practical value in confirming the diagnosis of cerebral echinococcal disease (22,28). The results of both of Casoni and Weinberg tests were also negative in our patient. CT scan and MRI are excellent techniques to diagnose and localize the lesions (1,5,6,18,23,24,27,40).

**Table I.** Summary of the previously reported primary multiple cerebral hydatid cysts.

| Authors and year | Age, sex | Localisation | Treatment | Outcome |
|---|---|---|---|---|
| Sharma et al. (34) 1982 | 9, F | 5 cysts in the right supratentorial region | Surgery | Good |
| Todorow et al. (37) 1988 | 47, M | 8 cysts in the frontal, temporal, parieto-occipital regions | Albendazole | Good |
| Paşaoğlu et al. (32) 1989 | 15, M | 3 cysts in the left fronto-parietal and 1 cyst in the left occipital regions | Surgery | Good |
| İplikcioğlu et al. (19) 1989 | 7, F | 3 cysts in the right frontal, 2 cysts in the left occipital, 1 cyst in the left frontal regions | Surgery | Good |
| Çataltepe et al. (10) 1991 | 8, M | 2 cysts in both parieto-occipital regions | Surgery | Good |
| Gupta et al. (18) 1991 | 18, M | Multiple cysts in both cerebral and left cerebellar hemispheres | Surgery | Died |
| Nurchi et al. (30) 1992 | 9, M | 30 cysts in the right parietal and occipital regions | Surgery | Good |
| Bilge et al. (4) 1993 | 37, M | 2 cysts in the left fronto-parietal and occipital regions | Surgery | Good |
| Martin et al. (26) 1996 | 69, F | Multiple cysts in both cerebellar and temporal regions | Albendazole | Died |
| Mancuso et al. (25) 1997 | 62, M | 1 cyst in the right frontal and 1 cyst in the left frontal regions | Surgery | Good |
| Baysefer et al. (3) 1998 | 20, M | 20 cysts in the left fronto-parietal regions | Surgery | Good |
| Popli et al. (33) 1998 | 20, M | Multiple cysts in the left temporo-parietal region | Surgery+Albendazole | Good |
| Özkan et al. (31) 2001 | 8, M | More than 25 cysts in the left temporo-parieto-occipital region | Surgery | Good |
| Nowak et al. (29) 2002 | 46, F | 2 cysts in the cerebral and cerebellar regions | Albendazole | Good |
| Karadag et al. (20) 2004 | 45, F | 2 cysts in the right parietal region | Surgery | Good |
| Yurt et al. (39) 2007 | 19, F | 24 cysts in both cerebral hemispheres | Surgery+Albendazole | Good |
| Erkutlu et al. (13) 2008 | 15, M | 2 cysts in the right parietal and occipital regions | Surgery | Good |
| *Present case* | 15, M | 19 hydatid cysts in the right parieto-occipital region | Surgery | Died |

Surgery is the standard and most effective treatment for intracranial hydatid cysts. The aim of surgery is to remove the cysts without rupture. The preoperative diagnosis is very important both in planning the surgery and taking the measures against spillage of daughter cysts and scolices at surgery. Rupture is associated with the well-recognized problems of anaphylaxis, meningitis, or local recurrence from spillage of the cyst contents (1,8,17,22).

The cavity remaining after the removal of large space-occupying cysts may cause serious complications, such as cortical collapse, cerebral edema, hyperpyrexia, cardiorespiratory failure or subdural hematomas in the early postoperative days. The development of subdural collections and porencephalic cyst has also been reported in the late postoperative days (15,36). The mortality rate of cortical collapse has been reported to be as high as 7% (11). Unfortunately the pressed and displaced brain parenchyme of our patient did not return to its original position. The patient's neurological status did not improve and he died.

Nevertheless, hydatid cyst is a benign lesion. Appropriate and timely management is mandatory for reducing mortality and morbidity.

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