

# Multiple Localization of Intracranial Cyst Hydatid in an Adult Patient: Case Report\*

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## ABSTRACT

*Cyst hydatid is a parasitic disease caused by Echinococcus larvae, seen frequently in countries where livestock farming is commonly practiced. Involvement of cyst hydatid in the central nervous system is rare. Multiple intracranial involvement with both supratentorial and infratentorial localizations is rare in adults. In this paper, we present an adult case with cyst hydatid with multiple localizations and a significant clinical picture.*

**Key words:** Cyst hydatid, Echinococcus granulosus, Central nervous system, Treatment

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## ÖZET

### Erişkin Hastada Multipl Yerleşimli İntrakraniyal Kist Hidatik: Olgu Sunumu

*Kist hidatik ekinokokkus larvalarının neden olduğu paziter bir hastalıktır ve hayvancılığın yaygın olarak yapıldığı ülkelerde sıklıkla görülür. Santral sinir sisteminde kist hidatik tutulumu nadirdir. Erişkinlerde hem supratentoriyal hem infratentoriyal multipl intrakraniyal tutulum nadirdir. Bu yazıda biz belirgin klinik tablosu olan erişkin bir olguda multipl yerleşimli bir kist hidatik sunduk.*

**Anahtar kelimeler:** Kist hidatik, Echinococcus granulosus, Santral sinir sistemi, Tedavi

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## INTRODUCTION

The type causing disease in humans is *Echinococcus granulosus*, or less frequently, *Echinococcus multilocularis*<sup>[1]</sup>. The main host is dogs, while intermediate hosts are domesticated mammals such as sheep and cows. Humans are generally infected by consuming foods contaminated with parasite eggs.

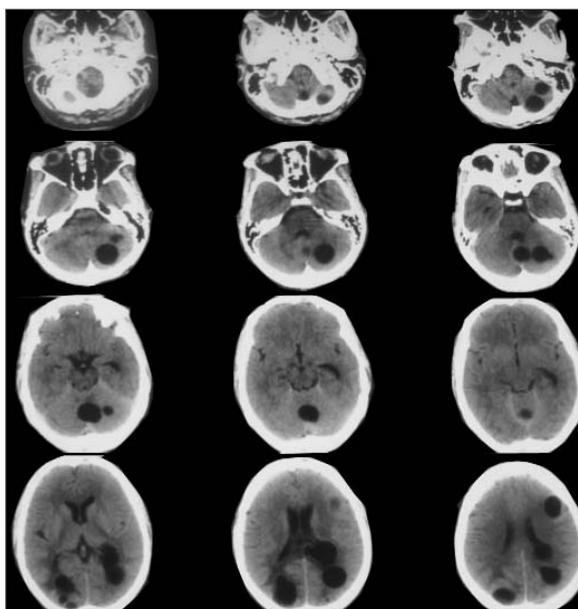
Eggs taken normally by the oral path are digested in the stomach, and then the released embryos are carried to the liver by the intestines and portal system. Rarely, some embryos are conveyed to the lungs through this path, and others to the systemic circulation and consequently to the brain<sup>[1,2]</sup>.

Involvement of cyst hydatid in the central nervous system is rare. They generally locate in the middle cerebral arterial region. Brain involvement occurs in 1-2% in *E. granulosus* infections<sup>[3]</sup>. Cranial localization is rare in adults. Approximately 75% of intracranial cyst hydatids are encountered in children<sup>[1,2,4]</sup>. Cerebral cyst hydatid is generally of single, round and unilocular appearance, and does not demonstrate serious symptoms until reaching a larger size. Serological tests as well as imaging methods, especially in cranial involvements, are of great value in the diagnosis of cyst hydatid<sup>[5]</sup>. In this paper, we present an adult case with supratentorial and infratentorial multiple hydatid cysts with a significant clinical picture.

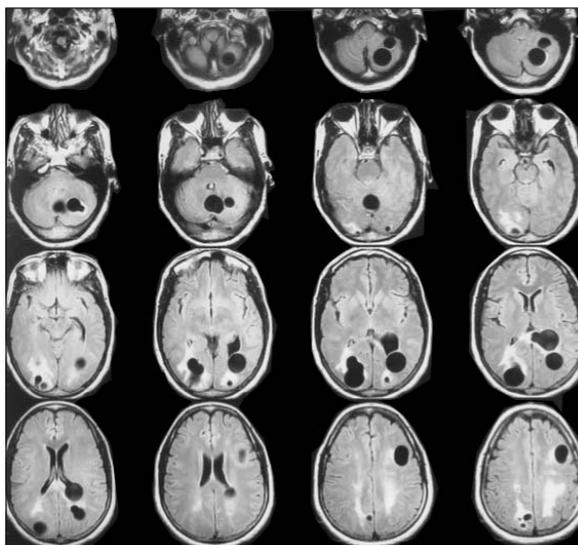
### CASE REPORT

A 40-years-old woman presented to our clinic with complaints of headache, nausea and visual impairment. Her history revealed that she worked in livestock breeding. Her visual impairment had increased in time, and she suffered from vomiting. She had experienced epileptic attacks on two occasions. Physical and neurological examinations revealed no significant findings other than slight stasis on fundoscopy. No pathologically significant findings were observed in laboratory tests. Computed tomography (CT) demonstrated hypodense cystic masses of various sizes with regular contours, with multiple localizations in the cerebrum and cerebellum (Figure 1). Cranial magnetic resonance imaging (MRI) revealed multiple supratentorial and infratentorial cystic lesions, the largest with a diameter of 2.5 cm, in the left cerebellar hemisphere and vermis, which were surrounded by slight edema. The lesions showed hypointense signal changes in T1-weighted images and hyperintense signal changes in T2-weighted sequences, suggesting cyst hydatid (Figure 2). Thorax and abdominal CT demonstrated a cystic hypodense lesion with regular contours in the spleen anterosuperiorly, having a hyperdense circumference of 23 x 27 mm. Cardiological examinations and echocardiography of the patient were in normal limits.

The patient was operated in two sessions. In the first operation, the cysts with supratentorial localization were removed with three separate craniotomies using Dowling's technique. The patient was operated



**Figure 1.** Computed tomography (CT) of cysts with multiple localizations in the cerebrum and cerebellum, showing hypodense cystic masses of various sizes with regular contours.



**Figure 2.** A 40-years-old woman with cerebral hydatid disease. Axial T1-weighted image shows well-defined, spherical, hypodense multiple cystic lesions in the supratentorial and infratentorial regions.

10 days later in order to remove infratentorial cysts. As the patient had no post-operative deficits, she was discharged under the supervision of medical treatment on the 5<sup>th</sup> post-operative day.

## DISCUSSION

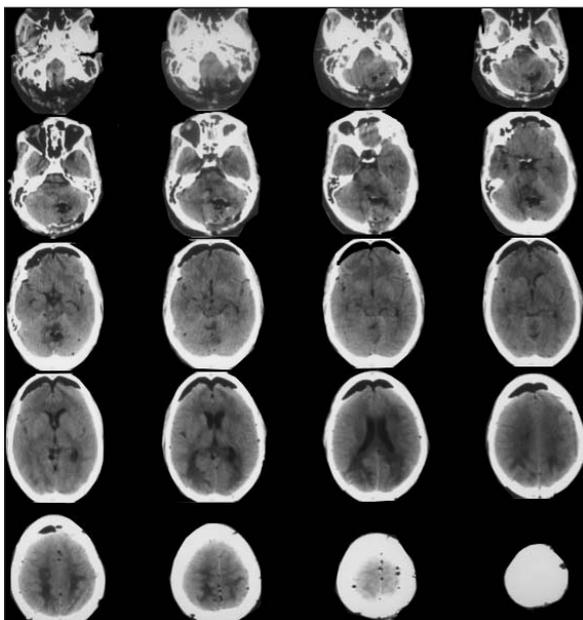
Cyst hydatid disease is most frequently observed in districts where livestock breeding is common. Cysts localized in various organs cause symptoms according to their mass formation effects. In the majority of the cases, the cyst is single and with large localization. Multiple localizations of the cysts are observed in 20-30% of the cases. The liver is the most frequently (70%) involved, followed by involvement of the lungs (20-30%). Involvements may also be observed in the brain, eyes, gastrointestinal system, and in bones, though rare<sup>[6,7]</sup>. Central nervous system involvement is rare in cyst hydatid cases, and this constitutes approximately 1.5-5.2% of all the cases. Clinical findings vary according to the localization, size and the distance of the cyst to the vital structure<sup>[8,9]</sup>. Apart from the pressure effect, allergic reactions may develop as a result of laceration of the cyst wall. Symptoms develop slowly, and neurological deficits are observed in the late period, generally due to the increase in intracranial pressure. Fatigue in extremities and visual impairment can also be observed depending on the cyst's location<sup>[10]</sup>. During diagnosis, papilledema is generally present. Lesions are often single; multiple lesions are very rare<sup>[9,11]</sup>. In our case, the patient presented with headache, nausea and papilledema with the diminution in vision, as is usually seen. Our case is important in that it is a multiple cyst case in an adult patient, which is rare in the literature. Intracranial hydatid cysts are more frequently located in the supratentorial compartment. Infratentorial localization is not common. Evliyaoğlu et al. showed that multiple cyst hydatid usually results from cardiac embolus<sup>[5]</sup>. Echocardiography remains the most reliable test in the diagnosis of cardiac involvement and location of cysts within the cardiovascular system. In our case, lack of cardiac pathology also showed that the involvement does not depend on cardiac emboli. Multiorgan involvement may have resulted in the multiple brain emboli in our case.

Diagnosis is achieved by clinical, laboratory and imaging methods. CT and MRI are valuable tools, along with serology. The cysts appear as well-defined, thin-walled, round cystic lesions, which are generally homogeneous, on CT and MRI. On MRI, cyst fluid has an isointense appearance as cerebral spinal fluid in T1 and T2-weighted images, while the cyst wall has a ring shape with low signal intensity both in

T1 and T2-weighted images<sup>[5,11]</sup>. T2-weighted images are superior to T1-weighted images due to low signal intensity, in contrast with high signal intensity, to demonstrate the cyst wall. Differential diagnosis should include abscess, cystic tumors, arachnoid cyst, and pencephalic cyst.

Cyst hydatid can be distinguished from abscess and cystic tumors by the absence of a contrasting membrane and by absence of mural nodules. Other cystic lesions are generally not spherical in shape<sup>[12,13]</sup>. Cerebral cyst hydatid can be seen as a solid, semisolid or multilocular cystic mass, and calcification and peripheral edema are frequent. Following intravenous contrasting agent, contrast involvement is seen around the cyst. CT and MRI, alone or together, are very successful in achieving cyst hydatid diagnosis, and are also a good means for treatment in the pre-operative period<sup>[5,11,14]</sup>. Our patient had multiple small cysts in the cerebrum and cerebellum. Cranial MRI showed well-defined, spherical, hypodense cystic lesions in the parietal, occipital, frontal, cerebellar, and ventricular regions. On contrast-enhanced T1-weighted MR images, we clearly identified the pericyst capsules. CT scans showed no calcification in the cysts. Our case is in accordance with the literature.

Medical and surgical treatment should be performed simultaneously. Several surgical methods have been employed, such as enucleation of the cyst without bursting it (using Dowling's technique), discharging cyst fluid and removal of the germinal layer followed by capitonnage, and marsupialization of the cyst. One of the most curative methods is to enucleate the cyst without bursting it by Dowling's technique, followed by rinsing of the operation cavity with hypertonic sodium chloride<sup>[10,15-17]</sup>. We performed Dowling's technique in our case. The Dowling technique, later improved by Arana-Iniguez and San Julian, has been widely used for the surgical treatment of hydatid cysts of the central nervous system<sup>[18]</sup>. The essential steps of this technique are the following: creation of a large skin and bone flap; careful handling during all operative steps to avoid monopolar coagulation; opening the atrophic cortex overlying the cyst over an area with a diameter no less than three quarters of the diameter of the cyst; and allowing the cyst to fall out by just lowering the head of the operating table and flushing warm saline between the cyst and surrounding brain<sup>[17,19]</sup>. Dowling's technique was performed to re-



**Figure 3.** Post-operative CT shows successful removal of all the cysts.

move multiple supratentorial and infratentorial cysts. In our case, removal of cysts in the supratentorial region was considered in the first operation to avoid tentorial herniation. On the other hand, since there was a risk of upward herniation in the supratentorial removal, we decided pressure in the posterior fossa might have been lower. The hydatid cysts were then totally excised. Removal of large and multiple cysts from the brain may be difficult, and should be performed carefully to avoid premature rupture of the cysts. In our case, post-operative CT demonstrated successful removal of all the cysts (Figure 3), but there is a chance that small cysts not seen on CT or MRI may be left behind untreated. Thus, close follow-up is necessary with detailed MRI. The patient was in good condition with no neurodeficit after the operation, and albendazole was recommended for 8-12 months. She was referred to the general surgery department for her abdominal localizations.

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