

Multiple and Bilateral Primary Brain Hydatid Cyst Dowling Technique is not always Appropriate (Case Report)

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ABSTRACT

Hydatid disease is a parasitic infestation caused by *Echinococcus granulosus*. Echinococcosis occurs worldwide and can affect multiple organs. Majority of the intracranial cysts are secondary and solitary. Bilateral and multiple primary cerebral cysts are uncommon and occur as a result of rupture of primary cerebral cyst or embolization from a ruptured peripheral cyst. Emergency surgical management of a 20-year-old man with multiple and bilateral primary

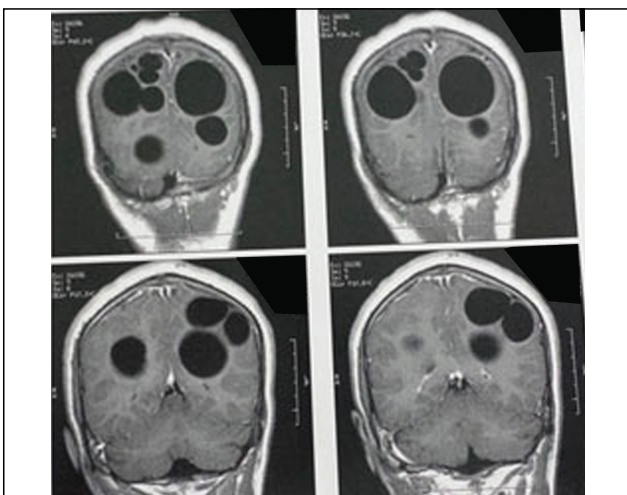
hydatid cysts are presented. Eleven cysts, which were symptomatic due to their mass effect, were surgically removed in two stages, followed up with medical treatment. The patient was admitted firstly, with blindness, headache, vomiting and frontal syndrome six months later. The patient deteriorated rapidly with signs of left temporal commitment. Postoperative outcome was satisfactory. In addition to the fact, that the presented case is an additional example for the rare primary multiple and bilateral cerebral hydatid cysts.

Keywords: Cerebral cyst, Echinococcosis, MRI

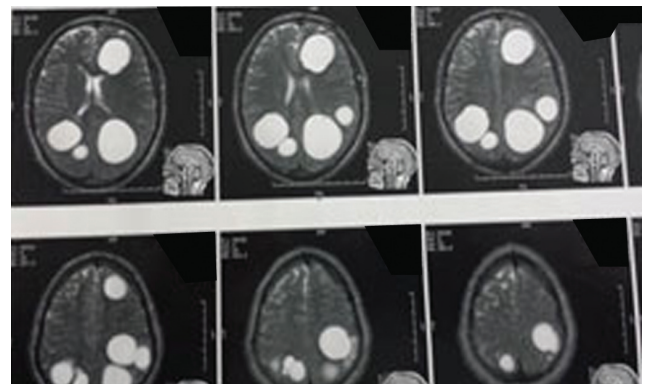
CASE REPORT

A 20-years-old man, the member of a family dealing with stockbreeding was admitted to the hospital with blindness, unremitting headache, vomiting and frontal syndrome. Neurological examination revealed bilateral papilledema without neurological deficit. Cranial MRI showed bilateral and multicystic lesions. Those lesions were isointense to CSF on all sequences and compressed lateral ventricle. The larger cyst was located in the left frontal lobe measuring 4x3x3 cm in size [Table/Fig-1&2]. The patient was scanned

to evaluate other possible organ involvements. Chest X-rays, abdominal ultrasonography and cardiac echo detected no findings suggestive of other systemic involvement of hydatid cyst disease. Additionally, serological test was very positive at 1560 UI/ml (normal < 160 UI/ml). Routine laboratory tests were within normal limits. The patient deteriorated rapidly with signs of left temporal commitment. The left cystic lesions were removed surgically to relieve the significant cyst-related mass effect. A relatively large frontal and occipital craniotomy were applied and cortical dissection was performed. Care was taken to avoid the rupture of the cysts during cortical dissection, but this was not possible for not worsening brain injury. The multicystic lesions consisting of 5 cysts was removed totally using puncture aspiration and continuous warm saline



[Table/Fig-1]: MRI T1 : Hypo intense bilateral and multiple brain cystic lesions.



[Table/Fig-2]: MRI T2 : hyper intense brain cystic lesions.

injection. The patient has evolved, with rapid recovery. One week later, six occipital cystic lesions in the other hemisphere were removed by the same technique. A total of 11 cysts were surgically removed. Histopathological examination confirmed hydatid disease caused by *Echinococcus granulosus*. Albendazole treatment was administered (10 mg/kg/day) for six months. Outside squellear blindness, no epileptic seizures, neurological deficits or sequel observed and follow-up radiological exams showed no evidence of hydatid disease.

DISCUSSION

Hydatid cyst disease is seen worldwide, but particularly in livestock producing countries. *Echinococcus granulosus* is endemic in some regions of the world including Mediterranean countries, the Middle East and Central Europe. The incidence of the disease in endemic regions ranges from 0.00003 to 0.0005 [1]. Cerebral involvement is approximately 1-3% of patients infected with the parasite, and this portion represents only 2% of all intracranial space-occupying lesions [2]. However, cerebral hydatid cysts are generally seen as single lesions and multiple cerebral cystic echinococcosis is a seldom entity in the setting of hydatid cyst disease [3,4]. We report a new case of multiple and bilateral cerebral hydatid cyst to discuss the save technique.

Hydatid cyst of the brain accounts for 1–3% of all Echinococcal infections [1]. Usually these are seen in the pediatric population and in the supratentorial compartment [5]. Multiple hydatid cysts of the brain are uncommon and may be either primary or secondary. Primary cerebral hydatidosis generally occur as a single lesion. Multiple cerebral cysts can sometime occur as a result of rupture of primary cerebral cyst or embolization from a ruptured peripheral cyst. The cyst may rupture as a result of trauma, during surgery or even spontaneously. Additionally, ingestion of multiple larvae can result in bilateral and multiple primary cerebral lesions [4]. The combination between cardiac and cerebral hydatid disease has been reported and evidence of embolization to the brain from cardiac lesions was documented [6]. However, very rarely a multiple larval intake may cause primary multiple cerebral hydatid cysts [7]. Clinical findings include headache, cranial nerve involvement, papilledema, focal neurological findings, cognitive impairment, convulsions and rarely hemichorea [3,7]. Immunodiagnosis can play an important complementary role. It is useful not only for primary diagnosis but also for follow up of patients after surgical or pharmacological treatment [8]. But the exact sensitivity and specificity of a given test for cerebral echinococcosis with or without extraneural involvement was not reported in the literature [9]. Serology was always negative in intracranial solitary lesions, but more sensitive in diagnosing cysts with multiple organs involvement, especially including liver [10]. Computerized tomography scans and magnetic

resonance characteristically show hydatid cysts as spherical, well-defined, non-enhancing cystic lesions disease [1]. The location is often superficial encouraging its enucleation without rupture. Multiple cysts pose various problems, not diagnostic but especially therapeutic. Surgery or isolate medical treatment [11].

Surgery is the treatment of choice for cerebral hydatid cysts. The aim of the surgery is the intact delivery of these cysts whenever possible [12]. Hydrodissection technique is the most useful method for intact delivery. For multiple cysts; an extra care should be taken to avoid the rupture during the extirpation of the cysts. But this technique can be dangerous and caused parenchymal damage in case of profound and eloquent location [10]. The rupture rate of hydatid cysts is higher in patients in whom the cyst is located deep in the parenchyma. Retractions that are performed to reach a cyst located in deep tissues of the brain as in our case, may cause temporary or permanent neurological deficits. Aspiration of the cystic contents through puncturing during the surgery for deep- seated cyst or cysts which located in critical areas is an alternative method [13]. Especially as multiple hydatid cysts resulting from the rupture of a primary cyst are infertile and have no broad capsule which permits the use of Dowling technique [14]. So the resultant risk of recurrence after their rupture is negligible. Our suggestion is to deliver multiple cysts one-by-one patiently with a small corticotomy. The largest one should be removed first, followed by the rest. In case of a rupture, careful suction must be performed in order to remove cyst content and surgical field must be irrigated with hypertonic saline solution. The surrounding parenchyma must be protected by cotton soaked hypertonic saline [15].

Medical treatment is highly recommended for patients with multiple cysts or deep-seated cysts, inoperable cysts and those who are not good candidates for surgery or may be suffering from recurrent cysts [10]. Albendazole may be administered for a number of reasons including to sterilize the cyst, decrease the risk of anaphylaxis, decrease the tension in cyst wall and reduce the rate of recurrence [15].

CONCLUSION

Multiple and bilateral cerebral hydatid cysts is exceptional. Hydrodissection technique is the most useful method for intact delivery, but this technique can be dangerous in case of profound and eloquent area. Aspiration of the cystic contents through puncturing is an alternative method associated to medical treatment.

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