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Primary multiple cerebral hydatid disease in a young patient with surgically-treated intracerebral haemorrhage. A case report

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Abstract

Introduction: Cerebral hydatid disease (CHD) is rare and the multiple-cystic variety is even rarer. In this paper, we report a case of multiple CHD and explore a possible link with a preceding spontaneous intracerebral haemorrhage (ICH).

Case presentation: A 27-year old gentleman with a history of surgically-evacuated, spontaneous ICH presented with severe headache, left-sided weakness - Medical Research Council (MRC) grade II - and recurrent tonic-clonic seizures, while on a full dose of anti-epileptic medication. Brain magnetic resonance imaging (MRI) scans showed multiple intra-axial cystic lesions in the right hemisphere. The cysts were removed intact using Dowling's technique through a large temporoparietal craniotomy. The surgery went uneventful and the patient recovered as expected. Post-operatively, a prophylactic course of albendazole (200 mg) was prescribed. On his one-year follow-up visit, the patient was symptom-free and his weakness had improved (left upper limb: MRC grade IV and full power of the left lower limb). The computed tomography (CT) scan showed no new findings.

Conclusion: Primary cerebral hydatid disease is rare and the multiple-cyst variety is even rare. In this case, a peculiar association with a surgically-treated ICH was explored with possible theories to suggest future research directions.

Introduction

Hydatid disease is a parasitic infection caused by an endemic parasite, Echinococcus Granulosis, mostly present in sheep-raising countries (1, 2). Tapeworm larvae develop in the intestinal tract then pass through the bloodstream to reach the liver, lungs, kidney, and brain (3).

Cerebral hydatid disease (CHD) comprises only 2% of all hydatid-cyst cases and is classified as "primary" or "secondary" (4). "Primary" CHD

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presents is rare and presents as a solitary single cyst surrounded by a broad capsule while the "secondary" type is characterized by multiple cerebral cysts that result from the rupture of a single cyst (1,5).

Preoperative diagnosis is necessary to avoid intra-operative cyst rupture. Cerebral hydatid cysts typically present with signs and symptoms related to raised intracranial pressure with or without focal neurological deficits depending on cyst size and location (5,6).

The authors report a case of a young patient with primary multiple cerebral hydatid disease and question its theoretical link with a prior, surgically-treated spontaneous ICH presenting at the age of 24.

Case report

A 27-years-old male presented with a single incidence of a generalized tonic-clonic seizure, headache, and vomiting. The patient had no history of head trauma. On examination, he had a left-sided weakness (MRC grade III) of both upper and lower limbs. Initial cranial CT scan (figure 1A) revealed an acute intracerebral hemorrhage (ICH) in the right parietal lobe. The hematoma was evacuated through the right parietal parasagittal approach. The postoperative CT scan revealed complete evacuation of the hematoma (figure 1B) and the post-operative course went uneventful. One month later, the patient's symptoms had resolved and the CT scan revealed no new findings (figure 1C).

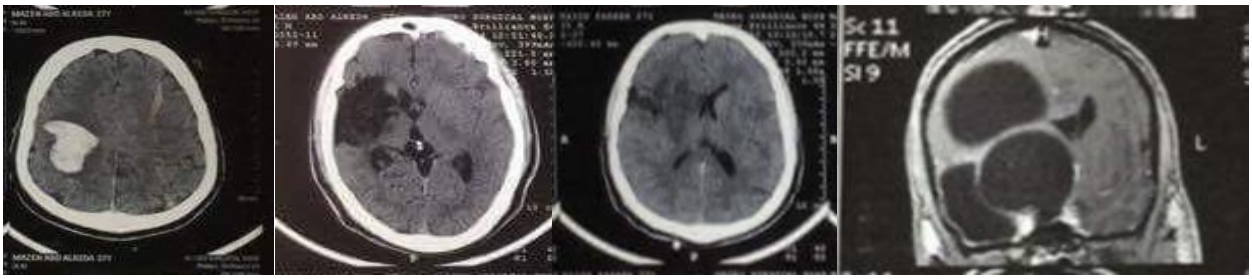

Figure 1. (A) Pre-operative CT scan (axial section), showing a well-defined, intracerebral hyperdensity representing intracerebral hemorrhage (ICH). (B) Post-operative cerebral CT scan (axial section) (C) Follow-up cerebral CT scan axial section shows no intracranial abnormalities. (D) FLAIR MRI coronal section showing a cystic lesion with a thin, hypointense marginal membrane with a marked mass effect causing contralateral displacement of midline structures.

Three years later, the patient presented with severe headache, left-sided weakness (MRC grade II), and recurrent tonic-clonic seizures, not responding to a full dose of anti-epileptic medication (carbamazepine 400mg bid). Cerebral CT and MRI scans (figure 1D) showed multiple intra-axial cystic lesions in the right hemisphere (figure 1). The cysts were removed intact using Dowling's technique through a large temporoparietal craniotomy. The surgery went uneventful. Post-operatively, the patient was started on a prophylactic course of albendazole (200 mg). His left-sided weakness remained the same (MRC grade 2). During his one-year follow-up visit, the patient was symptom-free and his weakness had improved (left upper limb: MRC grade IV and full power of the left lower limb). The CT scan showed no new intracranial pathology.

Discussion

Echinococcosis is an endemic infection in Mediterranean countries and Middle Eastern including Iraq. CHD is rare in comparison to other organ involvement, particularly the liver and lungs (7). Cerebral hydatid disease accounts for 0.05% of all cerebral mass lesions and is 2-3 times more common in children (3,8,9).

CHD can be primary or secondary. Primary CHD is the product of direct larval infestation in the brain without evidence of extracerebral disease. Secondary CHD occurs when the larvae exit the capillary beds of the lungs or liver to systemic circulation until they settle in the brain (10, 11). Primary CHD is uncommon, accounting for 1-2 percent of all Echinococcus granulosus infections, and multiple primary CHDs are even rarer (11-13).

Cerebral hydatid cysts are benign, slowly growing lesions with an annual growth rate of 1.5 to 10 cm and they can reach a considerable size before manifesting clinically. Headache, vomiting, symptoms of elevated intracranial pressure (ICP) are

some of the common presentations of cerebral hydatidosis (14). Other presentations are dictated by cyst size and location and may include hemiparesis, seizures, visual field disturbances, and gait disorders. Symptoms of raised ICP tend to predominate in the pediatric populations, while adults are more likely to present with focal neurological deficits and seizures (15,16). Cerebral hydatid cysts are most frequently located in the territory of the middle cerebral artery, commonly in the parietal lobe. The cysts are often found in the supratentorial regions and only rarely in the posterior cranial fossa, ventricles, or orbit (17).

Surgical excision of cysts without disruption of the capsule remains the standard management of CHD. A variety of surgical techniques have been developed to prevent cystic rupture. The most widely accepted method is Dowling's technique, in which the cyst is removed intact by injecting saline between the cyst wall-brain interface. This is made possible by the minimal adhesions that surround the cyst wall (18). Other techniques include cyst puncture and aspiration before removal or using cortical incision over the cyst, and ejection of the cyst by air insufflation of the contralateral ventricle (19). Possible postoperative complications include subdural effusions, obstructive hydrocephalus, and cortical collapse. In our case, the large spaces left behind after removing the three cysts were filled with isotonic saline prior to dural closure, in an attempt to reduce the risk of cortical collapse which carries a mortality of 7% (1,15). The follow-up CT scan showed a decrease in cavity size and an increase in brain tissue.

Medical treatment of hydatid cyst is required in cases of cyst rupture, recurrence, or systemic hydatid disease. It may also be used pre or post-operatively to shrink the cysts and prevent a recurrence, respectively (19-22). Our patient presented with three large cysts of different sizes and thus albendazole was prescribed post-operatively.

Whether this patient's past history of surgically evacuated ICH is related to CHD deserves further analysis. Although the sequence of events may be coincidental, the presence of spontaneous ICH in an otherwise healthy gentleman hints to a potential underlying connection. One theory is that surgical leukomalacia encouraged cystic development, particularly that the two pathologies are located in the same hemisphere. One other hypothesis is that the ICH provided an earlier manifestation of an occult CHD. Hemorrhagic CHD is, however, an uncommon occurrence. Another explanation is that this patient is a case of iatrogenic hydatidosis, which warrants institutional inquiry. In either case, we felt that documenting this case and discussing the possibility of association could direct further research efforts to better understand the etiology and presentation spectrum of CHD.

Conclusion

Primary cerebral hydatid disease is rare and the multiple-cyst variety is even rare. In this case, a peculiar association with a surgically-treated ICH was explored with possible theories to suggest future research directions.

Abbreviations

CHD: Cerebral Hydatid disease; ICH; intracerebral hemorrhage; MRC:

Medical Research Council; MRI: Magnetic Resonance Imaging; CT: Computed tomography;

ICP: Intracranial pressure.

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