Intra cranial hydatid cyst: A case report of total cyst extirpation and review of surgical technique

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ABSTRACT

Hydatid cysts commonly affect liver and lung but it can also affect the brain in rare cases. We report a case of 22 year female with history of headache for one and half years. Intracranial hydatid cyst was diagnosed on computed tomography scan and magnetic resonance imaging. The cyst was delivered without rupture using hydrostatic dissection followed by post-operative anthelminthic medication. Surgery remains to be the standard management. Amongst the surgical techniques described, Dowling's technique is the most acceptable. However, care must be taken in to avoid rupture of the cyst peroperatively which can result in subsequent complications and recurrence. Albendazole and corticosteroids can be used as adjunct to surgical treatment in selective cases.

Key words: Antihelminthic medication, craniotomy, hydrostatic dissection, intracranial hydatid cyst

Introduction

Hydatid cyst is an emerging zoonotic parasitic disease throughout the world.^[1] It is more prevalent in Australia, New Zealand, South America, Russia, France, China, India, Middle East and Mediterranean countries.^[2,3] Liver and lungs are the most frequently involved organs.^[4-6] However, it has been reported to involve brain,^[3,4,6-9] heart,^[4,5,10] kidneys,^[5] spinal cord,^[3,10] orbit,^[11] and spermatic cord.^[11] In the brain, it most frequently involves supratentorial region, mainly in the territory of the middle cerebral artery within the parietal lobe.^[12] Surgery remains the treatment of choice; nonetheless, careful removal of the lesion is of considerable importance; otherwise fatal complications are inevitable.^[13,14]

Case Report

A 22-year-old unmarried female presented to our outpatient department with one and half year history of

Access this article online	
Quick Response Code:	
	Website: www.ruralneuropractice.com
	DOI: 10.4103/0976-3147.116445

headache of mild to moderate intensity, dull in nature, with no variation with time. She had no complaint of vomiting, tinnitus, hearing or vision loss. There was no history of head injury. Fundoscopy revealed normal fundi. There were no lateralizing or localizing signs. Laboratory data showed mild leucocytosis without significant eosinophilia. Her liver function tests were within normal limits. Plain chest X-ray and ultrasound scan of the abdomen and pelvis did not show any abnormality. CT scan of brain demonstrated a single large, spherical, well-defined, thin-walled homogenous, iso-dense cyst in the frontoparietal region, with non-enhancing wall [Figures 1a and b]. This cyst was causing a midline shift of 1 cm, as well as dilatation of ventricles. MRI showed unilocular isointense cyst without surrounding edema [Figure 2]. Hence the diagnosis of a hydatid cyst was made. The patient underwent right frontotemporoparietal craniotomy. Following removal of bone flap and opening the dura, a small corticotomy of 1.5 cm was made in the frontal lobe through which the cyst was identified. A 10 French drainage tube was passed between the cortex and the cyst wall. Normal saline was carefully pushed in through this tube (Dowling's technique) to deliver the cyst measuring 5.5×5.8 cm² [Figure 3]. A hydatid cyst was confirmed on histopathology. A CT scan after 48 hours of surgery showed a large cerebrospinal fluid (CSF) filled space without any residual cyst [Figure 4]. Post-operatively, Albendazole 10 mg/kg twice daily was started and continued for four months. The patient

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was discharged on third post-operative day with close follow-up. On subsequent follow up visits the patient had no headache or seizures.

Discussion

Hydatid cyst is the larval form of the tapeworm *Echinococcus granulosus* which causes cystic echinococcosis, or *Echinococcus multilocularis* which causes alveolar echinococcosis; former being more common.^[15] It is an important zoonotic disease which is widespread throughout the world.^[11] The life cycle of this worm requires a definitive (a carnivore) and an intermediate (an herbivore) host. Humans act as an intermediary host and a dead end in its life cycle. It is transferred by ingestion of the scolex or eggs, or by direct contact with dog faeces.^[14] Only 1-2% of the hydatid cyst reaches the brain after passing through the liver and the

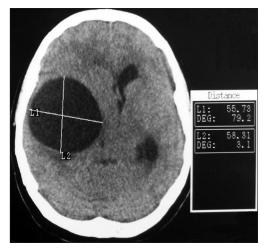


Figure 1a: CT scan brain plain showing large cystic lesion in right temporoparietal region causing effacement of the ventricle and midline shift

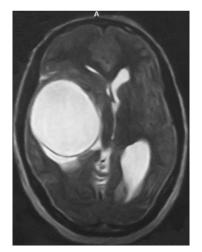


Figure 2: MRI T2W image of brain showing a CSF intensity cystic lesion in right temporoparietal region

lungs. It accounts for only 1-2% of all intracranial space occupying lesions.^[15,16] About two-third of the cases occurs in pediatric population.^[17]

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.^[18-20] The cyst may rupture as a result of trauma, during surgery or even spontaneously. Additionally, ingestion of multiple larvae can result in multiple primary cerebral lesions.^[21]

The symptoms of hydatid cyst depend on the location and the size of the cyst. They are either because of local compression or as a result of raised ICP. Headache is the commonest symptom while others include nausea, vomiting, vertigo, ataxia, seizures, lethargy, visual disturbances and cranial nerve involvement along with

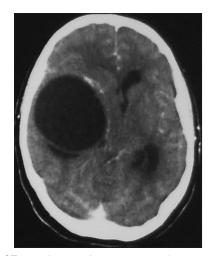


Figure 1b: CT scan brain with contrast revealing no enhancement of the lesion

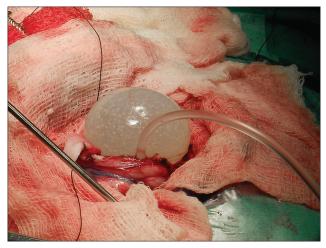


Figure 3: Peroperative photograph of the patient showing normal saline being pushed via a catheter between the cyst wall and brain parenchyma in order to deliver the cyst unruptured

focal neurological deficit depending on the location and size of the cyst.^[22] Cerebellar symptoms may arise when the cyst is located in posterior fossa.^[8,22] The pathological diagnosis of the disease is mostly based on detection of anti-*Echinococcus* antibodies immunoglobulin G (IgG) using indirect hem agglutination test or Enzyme-linked Immunosorbent Assay. The antibody titers are usually low or absent in cerebral hydatid cyst. However, there is no specific serologic test for cystic echinococcosis antibody detection.

MRI and CT scan are the investigations of choice for radiological diagnosis with MRI being superior for surgical planning.^[4,21] A CT scan characteristically shows hydatid cyst as a well circumscribed spherical or ovoid, hypodense, non-enhancing cystic lesion with no pericystic edema.^[23] On MRI it appears hypointense on T-1 weighted image and hyperintense with a hypointense halo around the cyst on T-2 weighted image.^[24] Fine peripheral enhancement along with perilesional edema may be seen in the presence of active inflammation.^[18,24] There is rarely any evidence of calcification.^[23] However, Alveolar echinococcosis appears as solid cum cystic mass and is usually associated with contrast enhancement, perilesional edema and calcification.[25] Certain features of hydatid cyst which impose a challenge for a surgeon include minimal neurological deficits despite the presence of a large cyst, diagnosis at the stage of fairly large cyst, thin wall of the cyst, and deep or periventricular location of the cyst.^[26] Surgical extirpation remains to be the best treatment option worldwide. Dowling technique is the most widely acceptable surgical technique. This emphasizes on atraumatic delivery of the un-ruptured cyst. However, this may not be possible in cases where the cyst is infected resulting in diffuse adhesions with the surrounding brain parenchyma. The

important intra operative steps of Dowling's technique are; creation of large flap; avoiding monopolar cautery; corticotomy of no less than three quarters of the diameter of the cyst; flushing warm saline between the cyst and surrounding brain.^[27] However, in our patient a corticotomy of about 1/4 the diameter of the cyst was sufficient to deliver the cyst without rupture [Figure 5]. A fine 10 French tube was passed between the cyst and brain parenchyma through which warm saline was pushed. This is a crucial step for delivery of the un-ruptured cyst. Surgeon must be patient enough to rely on hydrostatic pressure of saline flushed into the cavity. Lowering the head end of the patient also facilitates the delivery of the cyst by gravity.^[26] Any traction on the cyst wall at any stage should be avoided as this may lead to intra operative rupture of the cyst, which is the most common and serious complication. This can lead to widespread parasite dissemination followed by severe inflammatory or anaphylactic responses and multiple local recurrence of the cyst.^[13,28] In cases of per-operative cyst rupture it is recommended that the cavity should be washed with hypertonic saline.^[4] However this should be avoided when there is any communication between the cyst and the ventricular system.^[29] In case of multiple cysts, the largest one should be addressed first, followed by the rest.^[26,30] Added precautions must be taken to avoid rupture of the cyst in these cases. This may include satisfactory exposure of each cyst and changing position of head for delivery of each cyst. Multiple cerebral hydatid cysts may be addressed via multi-stage surgery.^[22,30] Care must also be taken in cases where the cyst is located directly underneath the dura with no intervening brain parenchyma between the two. In these cases scrupulous dissection of the dura must be performed in order to avoid rupture of the cyst. Another condition which demands meticulous microsurgical technique is the presence of adhesions between the cyst and the brain parenchyma. All adhesions must be



Figure 4: Post-operative CT scan brain plain showing collection of CSF in place of the recently removed cyst. There is no pressure effect or midline shift



Figure 5: Per-operative picture showing comparison of the corticotomy with the diameter of the cyst

released before attempting the delivery of the cyst. The cavity that remains after the removal of large hydatid cyst may have life threatening consequences in the early post-operative period including; cortical collapse, cerebral edema, hyperpyrexia, cardiopulmonary arrest or subdural collection.^[7] Postoperative complications include seizure, subdural effusion, porencephalic cyst, hemorrhage, pneumocephlous, hydrocephalus and transient neurological deficits.^[8]

Although surgery remains to be the cornerstone of management of intracranial hydatid cyst, 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.^[30,31] Corticosteroid may have a place in the management as it helps reduce the pericystic edema. Our patient was prescribed Albendazole for four months while Decadron 4 mg intravenous (i.v.) thrice daily was used postoperatively for 3 days, which was then tapered. Additionally, prophylactic anticonvulsants were also prescribed.

Conclusion

Cerebral hydatid cyst is primarily a surgical illness requiring meticulous surgical skills. Albendazole and corticosteroids may act as an adjunct to surgical treatment; however, the importance of un-ruptured extirpation of the cyst can not be over stated.

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How to cite this article: Umerani MS, Abbas A, Sharif S. Intra cranial hydatid cyst: A case report of total cyst extirpation and review of surgical technique. J Neurosci Rural Pract 2013, 4(Suppl 1):s125-8. Source of Support: Nil. Conflict of Interest: None declared.