

Intracranial Hydatid Cyst: A Case Report of Three Cases

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ABSTRACT

A cerebral hydatid cyst is a less common condition as compared to hydatid diseases of liver and lungs. Surgical removal of the cyst without rupturing it should be the primary goal of surgery. We reported three cases of supratentorial cerebral hydatid cysts in this case-series. All of the cases belonged to three different age groups. One, 10 year old patient was previously operated at another hospital and presented with the recurrent disease. This patient was operated three times with a gap of six and nine months in-between surgeries. Currently, he is symptoms free for the last one year. Another, 40 year old patient was operated, 22 months back and is now disease free. Third case was of a 72 year old patient, who had a hepatic hydatid cyst as well. The relevant literature on the cerebral hydatid disease was reviewed and compared with our cases. The cerebral hydatid cyst should be included in the differential diagnosis of patients presenting with altered neurological status, especially in those who belongs to those regions where the disease is endemic. Total cyst removal without rupturing it should be the goal of surgery whenever possible. Postoperative treatment with Tablet Albendazole should be given in every case.

Keywords: Cerebral hydatid cyst; Echinococcus granulosus; Albendazole; Dowling-Orlando technique; Surgical extirpation.

INTRODUCTION

A cerebral hydatid cyst is a much less common condition as compared to hydatid diseases of the liver and lungs, and if not diagnosed in time, it can be resulted in significant morbidity and mortality. It mostly occurs due to a tape worm Echinococcus granulosus, which lives in the jejunum of dogs. People in many areas of Pakistan and Afghanistan are still living a nomadic life, which predispose them to this disease. Surgical removal of the cyst, without rupturing it, should be the primary goal of surgery. The disease hydatid causes from zoonotic parasites ¹by the infestation of the larvae of Taenia Echinococcus. The definite common host of Echinococcus is a dog. All mammals, especially cattle are considered intermittent hosts. The course of infection basically consists of two hosts, i.e., definitive and intermediate. Dogs are the most common definitive hosts while the

sheep are the most common intermediate host.²⁻⁷ When humans get infected from contaminated food from the dog feces, which contains ova of the parasite via feco-oral path, they also act as an intermediate role.² The disease has also incidence in Russia, South America, Australia, China, India, Mediterranean and Middle East. The most impacted organs are liver and lungs from this parasite.³⁻⁶ Through the route of gut mucosa and circulation through the portal circulation, the embryo used to filter by liver or by the lungs.⁷ The intracranial hydatid cysts accounts only 0.5 – 3.0 (%) of all hydatid diseases.⁶ It mostly impacts brain's supratentorial region mainly in the area of middle cerebral artery in the parietal lobe.⁵ We outlined three cases with intracranial hydatid cysts along with intracranial hydatid cysts, clinical manifestations and surgical outcomes. The patients were operated at the Neurosurgery Department at Lahore General Hospital.

CASE REPORTS

Case 1

A 10 year old patient presented in the outpatient department with two month history of progressive left sided body weakness and nausea. Examination showed a large craniotomy scar over right temporal-parietal-occipital region. There was a weakness of right upper limb (grade 3 power) and lower limb (grade 4 power) with House-Brackmann grade 4 left facial nerve palsy. Fundoscopy revealed bilateral papilledema. His past medical history revealed that he had been presented with headache, vomiting, fever and fits in Shaikh Zayed hospital Rahim Yar Khan on August, 2015. Initially, he was empirically treated with Antituberculous drugs for a few days, and later on CT scan (brain) and MRI (Figures 1, 2) brain were conducted. There was a multiloculated cystic lesion in right temporal-parietal-occipital region, which had a low intensity on T1WI and FLAIR sequence and high intensity on T2WI. Mass effect on ipsilateral lateral ventricle and midline shift of 1cm towards the lateral ventricle was seen. He was discharged on oral Albendazole, and operated five months later (January, 2016). The surgical notes of that procedure were not available when the patient presented to us. Biopsy findings were consistent with hydatid cyst.

The patient was admitted in our Neurosurgical

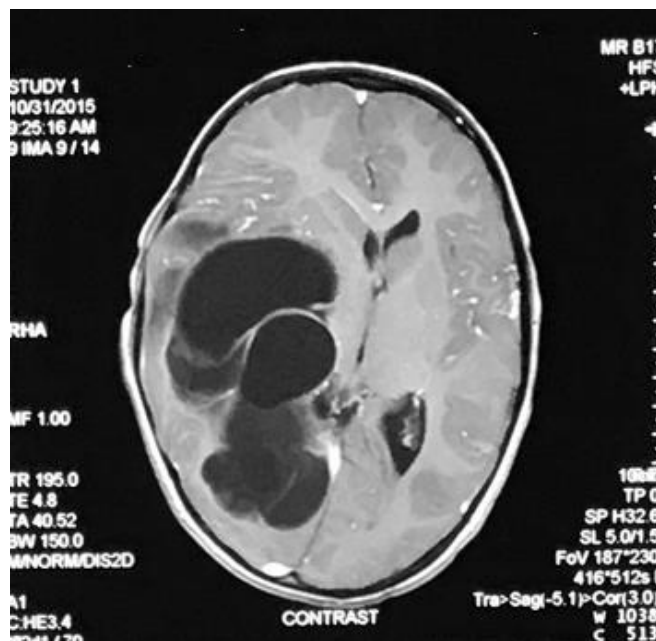


Fig. 1: MRI brain T1 weighted post-contrast image. Mild peripheral enhancement seen with hypo-intense cystic lesion.



Fig. 2: MRI T2WI showing hyperintense cystic lesion.

department. The CT scan brain and MRI brain were planned. Laboratory data showed no significant eosinophilia. Patient's LFT parameters were found normal. Moreover, abdomen ultrasound, plain chest X-ray and pelvis were also not diagnosed with any abnormalities. The CT scan brain showed large rounded intraaxial cystic lesions in right temporo-parietal region. MRI brain with contrast showed findings were consistent with recurrent hydatid cyst. Therefore, surgery was planned and patient was operated. The operation was done by using the same craniotomy incision. Dura mater was adherent to the underlying brain. Small superficial corticotomy was done and the cyst was identified. The cyst was adherent to the adjacent brain. Dowling's technique was tried, but due to the intense adhesions, it failed. Cyst wall was punctured and multiple 15 daughter cysts were removed one by one. The laminated membranes were partially removed, because of the concern of causing damage to normal brain. Post operatively, the left sided body weakness and facial palsy considerably improved. Histopathology report was consistent with the hydatid cyst, showing the laminated cyst wall with the evidence of protoscolices. The patient was discharged on Tablet Albendazole 200 mg twice daily and Tablet Praziquantel 40 mg/kg/week. On the subsequent two follow up visits, with three month interval, the patient's weakness had

greatly improved.

Eight months after the second surgery, the patient again presented with progressive left sided body weakness, headache and seizures. Weakness of limbs was more pronounced this time with grade 2 power in upper and lower limb. The CT scan and MRI brain showed a recurrent hydatid cyst at the same place. The patient was admitted and baseline investigations were done, which were unremarkable. Surgery was done in March, 2017, and operative findings were almost the same as in previous surgery. This time, hypertonic saline (10%) was used per-operatively. Laminated membranes were maximally excised in piecemeal due to the adhesions. Post-operatively, the patient had a slow recovery of limb power. He was discharged and referred to infectious disease specialist. Now he is on anti-seizure drug and has taken Albendazole for six months. Follow up visits (**Figure 3**) were regularly done and the patient has a grade 3 power in left upper limb and grade 4 powers in left lower limb. There were no headaches and seizures in the last one year. This first case was followed after 19 months of 3rd surgery. The patient had no recurrent symptoms, no headache and no papilledema. The power grade was grade 3 left upper limb and grade 4 in left lower limbs, which is same as observed in previous follow up visit.

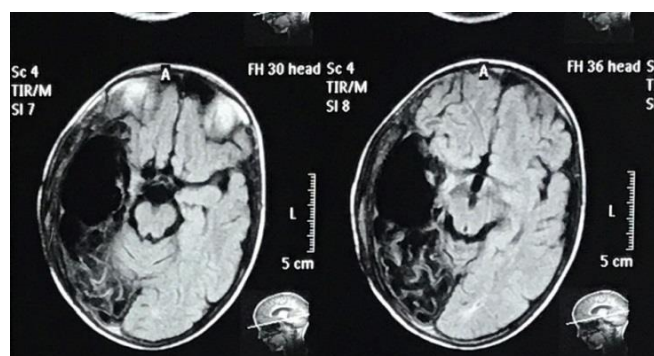


Fig. 3: Post-op MRI brain after 8 months of third surgery.

Case 2

A 40 year old male from rural area of Baluchistan was admitted in May, 2016 in the Neurosurgery department with headache for last three months as well as with vomiting and altered behavior for last 15 days. On physical examination he was disoriented with a bilateral papilledema. There was no history of head injury. The patient underwent cranial computed tomography (CT) and MRI (**Figures 4, 5**), which showed a single large intra-axial left parieto-occipital

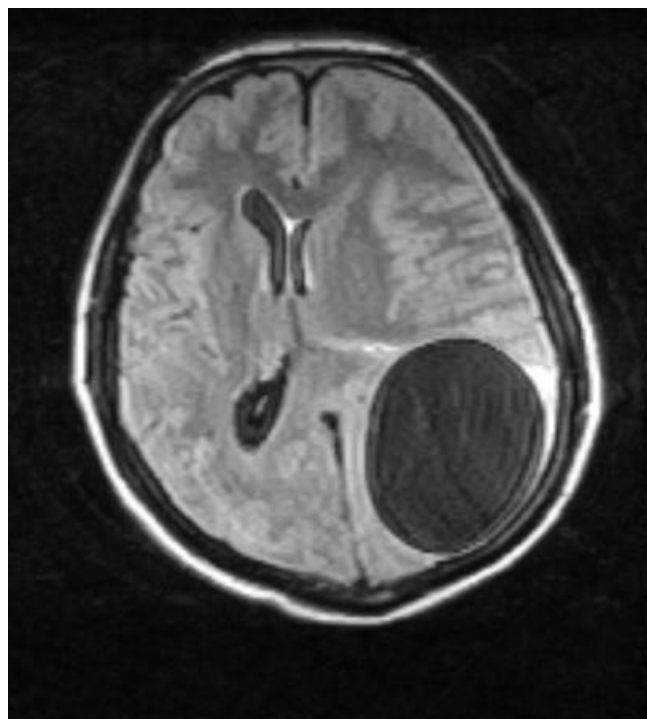


Fig. 4: T2 flair showing no perilesional edema

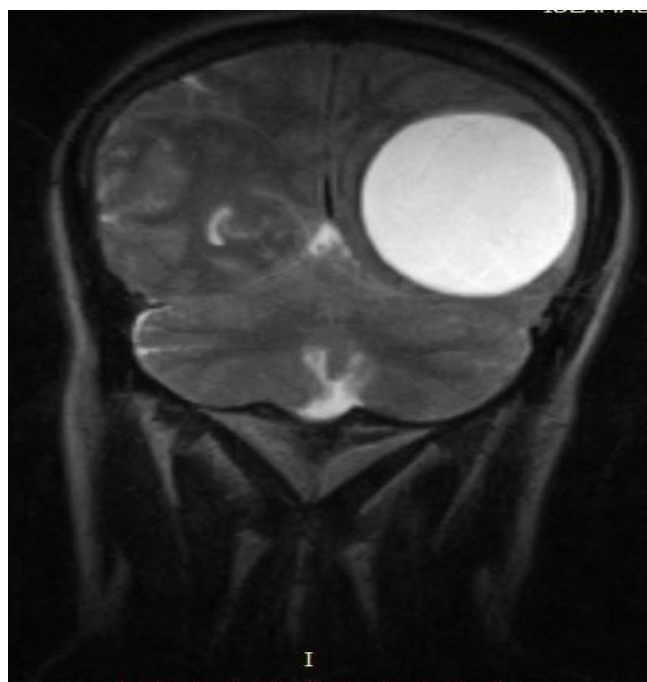


Fig. 5: Coronal T2WI of left occipital lobe hydatid cyst.

cyst causing a midline shift and ventricular compression. Hydatid cyst was diagnosed. The laboratory data showed no abnormality. Chest X-ray

and ultrasound of whole abdomen were unremarkable. The patient was operated via. left parieto-occipital craniotomy. Standard Dowling's technique was followed during the removal of cyst and, it got ruptured before being delivered to the receiving tray. Normal saline was used during the procedure. The histopathology findings were consistent with a hydatid cyst. The postoperative course of the patient was found uneventful, and discharged five days after surgery. Chemotherapy that included Albendazole 400 mg twice times daily was administered for three months. Twenty two months after the surgery, apart from mild memory disturbances, this patient's neurologic assessments were found normal. A CT scan of the brain after six months revealed no recurrent cyst (**Figure 6**). This second case was symptoms free after 29 months with mild memory disturbances which were present pre and post operatively.



Fig. 6: Post-op CT scan.

Case 3

A 72 year old male patient was referred to the Neurosurgery outpatient department with complaints of memory disturbances for three months. The patient had developed personality changes and fits for the last seven days. He was confused and disoriented to time, place, on examination. There was a bilateral papilledema. The cranial CT scan and MRI brain

(**Figures 8, 9**) showed multiple hydatid cysts occupying the right frontal lobe, causing a midline

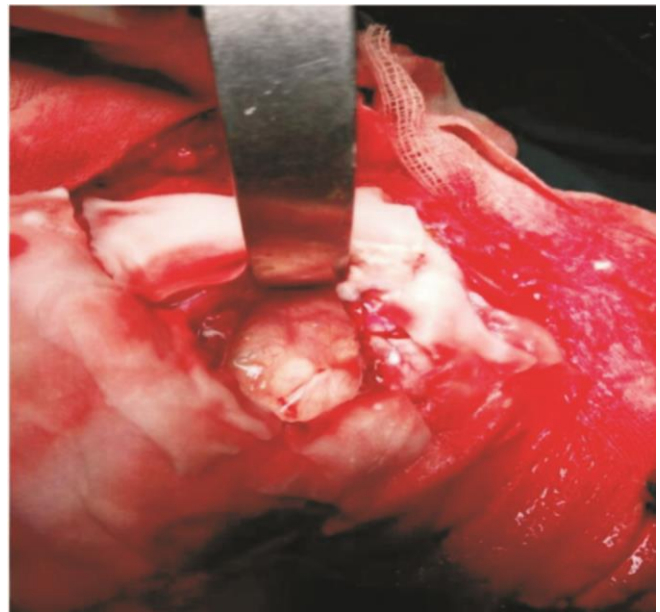


Fig. 7: Image showing an empty cavity after cysts removal.

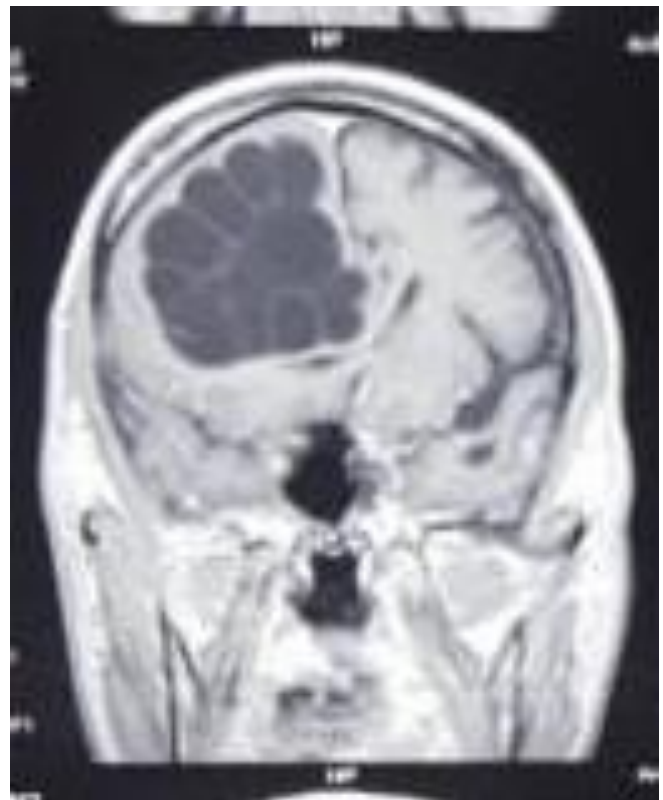


Fig. 8: Pre-op MRI brain T1WI coronal view showing multiple hydatid cysts in the right lobe.

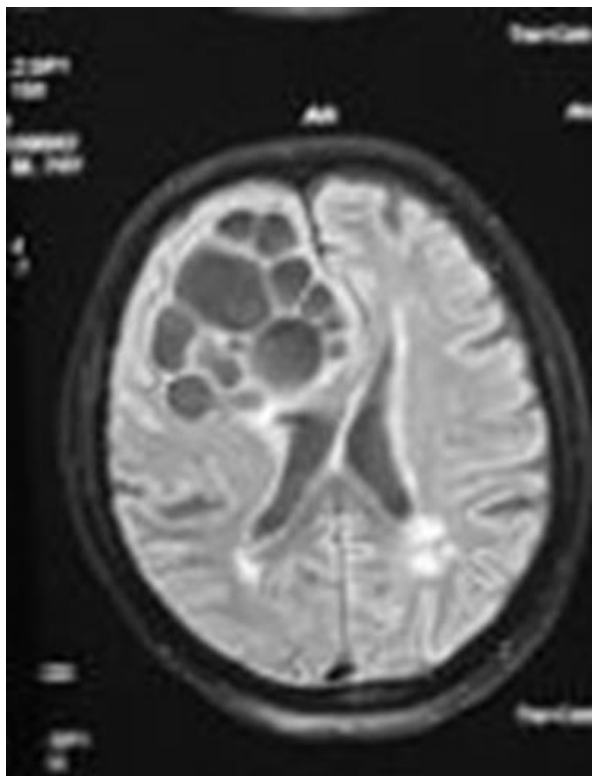


Fig. 9: Pre-op MRI T2 Flair showing no perilesional edema.

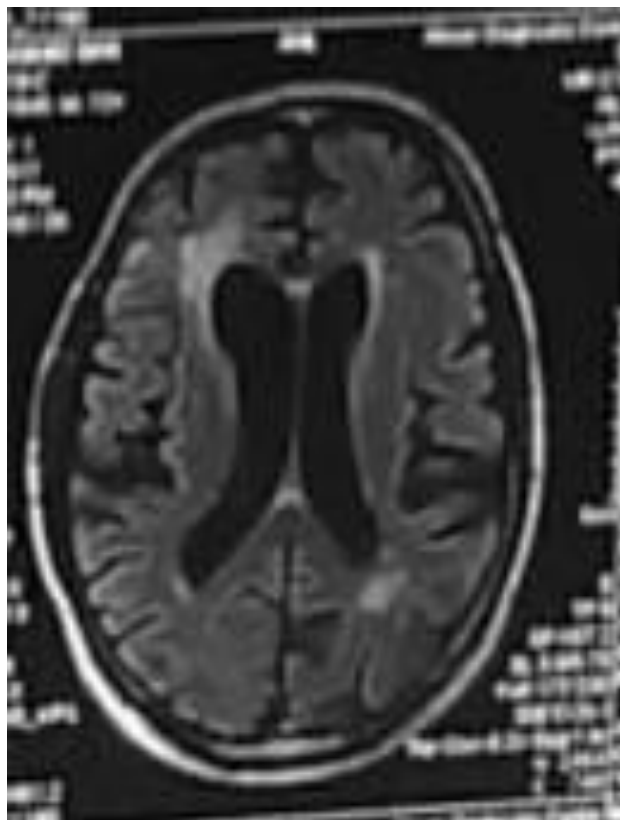


Fig. 11: Post-op T2WI axial view.



Fig. 10: Unruptured hydatid cysts.

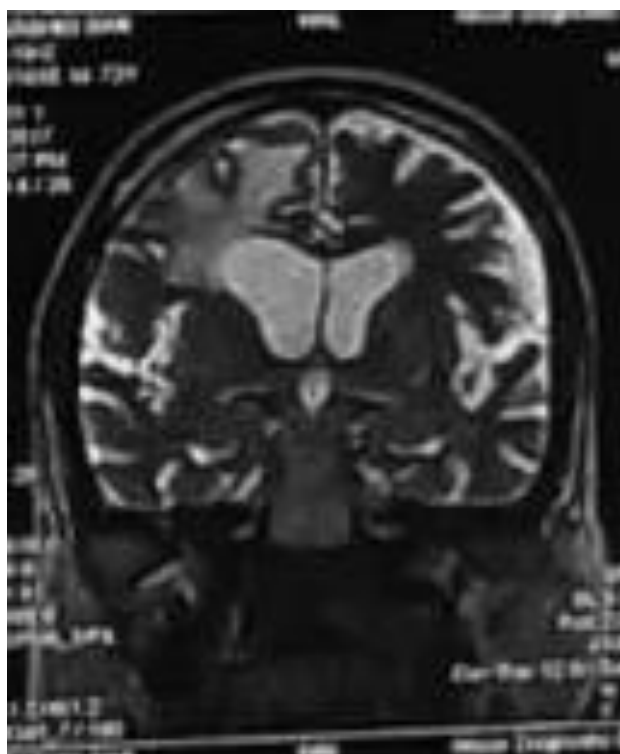


Fig. 12: Post-op T2WI coronal view.

shift and ventricular compression, hypo intense on T1-weighted sequences and hyper intense on T2-weighted sequences. No peripheral edema was noted around the cysts. Ultrasound liver showed a hydatid cyst in the liver. During the course of admission, he developed a left sided hemiparesis and aphasia. The patient was then instantly operated upon. A large craniotomy was performed. The 32 cysts were removed unruptured by 10% hypertonic saline irrigation. The pseudocyst capsule was then completely removed (**Figures 7, 10**). Postoperatively, the patient had a marked improvement in hemiparesis and higher mental status. A postoperative follow-up MRI brain (**Figures 11, 12**) revealed cyst-free intracranial content. Chemotherapy with Albendazole was continued and the patient was referred to the General Surgery department for the management of hepatic hydatid cysts, where he was operated. One year after surgery, he is well and free of disease. The third case was followed at 17th month postoperatively and is symptom free.

DISCUSSION

Echinococcosis is known as a hydatid disease and a hydatid cyst is practically a lethal disease that impact animals, cattle and humans.⁶ The disease spreads through some local and cultural practices and lifestyles. People in many areas of Pakistan and Afghanistan are still practicing nomadic life. Pakistan is an agricultural country, and folks used to have close contact with their livestock. Slaughtering practices at home, is still practiced widely in our region. The dog is fed with the internal organs of the slaughtered animal, which are suspected to have infections. The feces of the infected dog can contaminate the food, water and soil, which lead the initiation of the parasite's life-cycle. Veterinary services are improving in Pakistan and they are educating the masses to use anthelmintic drugs both for live stocks and dogs. Out of the three cases we reported, one belonged to Afghanistan and one belonged to Baluchistan. A third patient belonged to Punjab and he was a frequent visitor to national and international stations for media coverage. All of the three patients had dog pets in their homes.

Cerebral hydatid cysts are of two types, i.e., primary and secondary. Primary cyst is caused by those embryos that are not filtered by the liver and lungs. These are of the most common type and in most patients, they are solitary. They are fertile, because they brood capsule and scolices.⁷ Secondary cysts are

often multiple and may cause from the embolization of a ruptured primary cyst in other organs. They are non-fertile because of the absence of brood capsule and scolices. The histopathology reports of all of our cases were consistent with the primary types of the cyst. Only, 0.5-3.0 (%) of the hydatid cyst reach the brain and accounts for only 1-2% of all intracranial space occupying lesions. The signs and symptoms are nonspecific and mostly are due to the raised ICP (intracranial pressure). The most common ones are headache, papilledema, and vomiting. Focal symptoms like hemiparesis, seizures, gait, or visual deterioration can be observed, depends on the size/location of the lesion. They can be seen in any part of the brain, but are usually supratentorial and located in the middle cerebral artery territory, most commonly the parietal lobe.⁸ It is a slow growing benign lesion and literature reports a growth rate of 1-10 cm. In the pediatric age group, it can reach an enormous size due to an elastic nature of the skull.

Although, the diagnosis of hydatid cyst is pathological, a radiological investigation is very helpful in characterizing the hydatid cyst preoperatively. The initial investigation usually performed in cases of cerebral involvement on the CT scan (brain), which shows a spherical cystic lesion with well-defined borders. The MRI appears to be superior to the CT in the detection of rim enhancement and surrounding edema.⁹ On MRI, these lesions have a low intensity on T1WI and high intensity on T2WI. There is no or minimal perilesional edema, if significant then indicates to rupture or secondary infection. The differential form, which the hydatid cyst has to be distinguished are epidermoid cyst, cystic astrocytoma, arachnoid cyst, cystic granuloma and pyogenic abscess. Surgical extirpation for intracranial hydatid cysts is considered a gold standard treatment option. Dowling-Orlando technique is a recommended procedure. The basic rules of the technique include a wide craniotomy, ignoring monopolar cautery, corticotomy of no less than 3/4 of cyst's diameter, flushing warm saline between the cysts and surrounding the brain. In our cases, this technique was successful in two out of three cases and failed in one case of tight adhesions between brain and cyst. Therefore, in that case, the cyst was ruptured intentionally and aspirated, after that multiple cysts were removed. Few complications that may occur after excision of large cysts are cerebral edema, hyperpyrexia, cardiorespiratory failure, subdural collection, and the development of a porencephalic

cyst.¹⁰ Rupture is linked with “anaphylaxis, meningitis, or local recurrence from spillage of the cyst content”.¹⁰ Recurrence and meningitis were seen in one of our case, which was operated three times in total. The other two cases are disease free till now.

In order to control Echinococcosis in dogs and hydatid disease in cattle, a drug monitoring on a monthly basis should be conducted¹¹. The control would be successful when the measures will be applied on whole community. It includes following general precautions. Deworming of dogs that take an uncooked sheep viscera. The dogs should be given Praziquantel after every six weeks. There is a need a proper disposal of sheep viscera. The expulsion of stray dogs. Ensuring a proper hygiene if keeping pet dogs or sheep dogs as a livestock guardian.¹² Three effective and safe products are available against Echinococcus in dogs, such as DRONCIT (Praziquantel), DRONTAL PLUS (Praziquantel + Pyrantel Pamoate+ Febentel) and CESTEX (Epsiprantel).¹²

CONCLUSION

Cerebral hydatid cyst should be included in the differential diagnosis of patients with altered neurological status, especially those belonging to regions where the disease is endemic. Total cyst removal without rupturing it should be the goal of surgery whenever possible. Postoperative treatment with Tab. Albendazole should be given in every case.

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