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**Soumaya Touzani, Brahim Bechri, Toufik Joulali,
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ABSTRACT

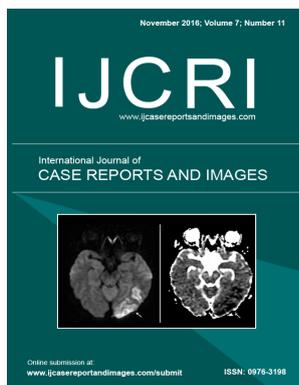
Introduction: Hydatid cyst disease in childhood is still a serious health problem in the rural areas of Morocco and other places where the parasite is endemic. Brain involvement rate is about 2% of the cases and is most commonly observed in children.

Case Report: We report the case of a five-year-old boy who presented with general weakness and vomiting before rapid deterioration of consciousness, requiring an urgent operation as there were signs of midbrain herniation with a cystic lesion in the left frontoparietal region at the Computed tomography scan. The patient was discharged after one week with optimal neurological status. Although cases of cerebral hydatid disease are quite common in literature, the urgent character of our case is rarely reported and will be discussed in the light of previous cases as well as cerebral hydatid disease management.

Conclusion: In case of emergencies, such as midbrain herniation with cystic lesions identified by CT scan, the possibility of hydatid cysts should be considered where the infection is endemic and the team should be prepared to remove the cyst without rupture.



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Keywords: Children, Dowling's technique, Hydatid cyst, Intracranial

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INTRODUCTION

Hydatid disease is an emerging zoonotic parasitic disease throughout the world. In Morocco, it is considered as a public health problem [1]. Brain involvement with hydatid disease occurs in 1–2% of all *Echinococcus granulosus* infections. It mainly affects children [2]. Through a rare pediatric emergency case of hydatid cyst brain, we remind the clinical and radiological presentation and surgical outcome of cranial hydatidosis.

CASE REPORT

A five-year-old boy was admitted to the emergency department, with an inaccurate history of vomiting and general weakness. On examination, we found a drowsy patient with a Glasgow Coma Scale (GCS) of 13/15 (E3 V4 M6), tachycardia at 135 beats/min and signs of moderate dehydration (5–10%). The patient was afebrile and his respiratory rate was 24/min. Capillary blood glucose was 1.5 g/dl. Intravenous rehydration was started after refitting and blood sampling for complete electrolytes. Before cranial computed tomography (CT) scan was taken, the patient had generalized epileptic seizures. The intravenous rehydration was interrupted and the convulsions have ceased after administration of midazolam. Serum electrolytes performed on admission were within normal limits. A neurological reassessment revealed anisocoria with dilatation of the left pupilla. Due to rapid deterioration of consciousness (GCS at 9), the patient was intubated and artificially ventilated. The CT scan (Figure 1) revealed a single large, spherical, well-defined cyst in the left frontoparietal region. This cystic structure caused a significant mass effect on the ventricular system and the midline with subfalcine herniation. A plain chest X-ray revealed a suspicious image of hydatid cyst of the left upper lung lobe. The examination afterwards revealed a history of headache and vomiting lasting for two months out of trauma, as well as rural background and contact with dogs. The diagnosis of a hydatid cyst complicated by intracranial hypertension (ICHT) was made. Because of the midbrain herniation signs, urgent operative intervention via a large craniotomy was performed under general anesthesia with one lung ventilation. Moderate hyperventilation (PaCO₂ 32–35 mmHg), high-dose of mannitol (1.5 g/kg), corticosteroids and anesthetic drugs were adopted for the perioperative management of ICHT. Anti-anaphylaxis measures were kept ready. Brain was well relaxed. By using the method of Dowling's technique of hydro-dissection, a large cyst was delivered with utmost care to avoid rupture and spillage (Figure 2). Valsalva maneuver was also applied and the patient's head was lowered to

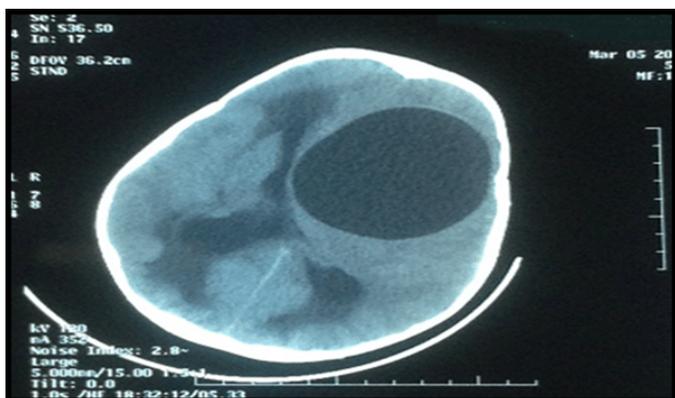


Figure 1: Computed tomography scan showing solitary cystic lesion located in the left frontoparietal region.

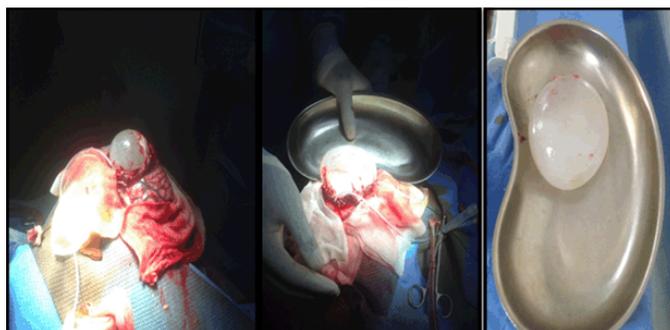


Figure 2: Perioperative appearance of the hydatid cyst during and after removal.

further ease the cyst removal. The dura was closed in a watertight fashion after filling the remaining cavity with 0.9% saline solution. The patient was extubated with uneventful recovery and shifted to the ICU for observation. The surveillance was assessed using the Glasgow Outcome Scale, the neurological examination and CT scan. He showed marked improvement in his mental status but kept a right hemiparesis. The pathology reports confirmed the hydatid nature of the cyst. The patient was placed on albendazole regimen 15 mg/kg/day and was discharged after one week with close follow-up and orientation to thoracic surgery.

DISCUSSION

Hydatid disease is endemic in Middle East, Mediterranean countries, South America, North Africa and Australia. Giant intracranial hydatid cyst (HC) is very rare, with reported incidence of 1–2% of all cases [1–3]. There was specific history of direct contact with dog in our patient. Cerebral hydatid cyst is more common in pediatric population, probably related to a patent ductus arteriosus. Intracranial hydatid cyst may be classified as primary or secondary [2]. In our case, larvae might have passed through the capillary filter of the liver and lungs, entered into the systemic circulation and reached the brain. Probable presence of lung hydatid cyst and normality of echocardiography performed afterwards consolidate this statement.

The cysts enlarge slowly, rates of approximately 1 cm per year are quoted, but this is variable and may be higher in children [2]. The development of symptoms is slow. Neurological deficits appear late, are often preceded by signs of increased intracranial pressure and may vary with the location of the cyst. In children and adolescents, the clinical picture usually includes the cardinal symptoms of increased intracranial pressure [2, 4]. Focal findings such as hemiparesis, speech disorders and hemianopsia, sometimes associated with epileptic seizures, and even coma are also reported in literature [5, 6]. In our case, the patient presented the most common symptoms (headache and vomiting) during two

months before developing coma and seizures 1 hour after admission.

The primary hydatid cysts mostly involve the territory of the middle cerebral artery, particularly in the parietal and frontal lobe, and generally cause significant ventricular distortion and midline shift, as in the present case [7]. The correct preoperative diagnosis, formerly arduous, can now be established by CT scan and MRI scan. The CT and MRI scan have tremendously increased diagnostic specificity and helped the surgeon to plan the cortical incision and accurately approach the lesion. We could not perform a cranial MRI scan prior to the operation due to the poor condition of the patient. He underwent an urgent operation, based on CT findings which showed hypodense cystic lesion that was sharply demarcated and without perifocal edema and fluid identical to cerebrospinal fluid. In literature review, there are four cases, to our knowledge, that received an urgent operation for a cerebral hydatid cyst [5, 6, 8, 9] with success in three cases.

The treatment of intracranial hydatid cyst is surgical. The aim of surgery is to remove the cyst in toto without rupture to prevent recurrence, chemical meningitis and anaphylactic reaction. A variety of surgical techniques is used but the preferred method remains the Dowling's technique of hydrodissection in which normal saline irrigation between the cyst and the surrounding brain parenchyma is used to deliver the cyst intact. This is possible because of minimal adhesions around the cyst wall. Location and multiplicity are the two main problems faced by surgeons in treating this disease [10]. The other main concern in hydatid cyst resection faced by the anesthesiologist is allergic reactions after the rupture that can be range from mild hypersensitivity to fatal anaphylactic shock [11]. A combination of thorough preoperative evaluation, intensive monitoring and a good technique that helps maintain stable intraoperative hemodynamics helps in achieving a successful outcome in such patients.

Recurrence remains a major concern, which is managed by both antihelminthic chemotherapy and surgery. In a study conducted by Ciurea et al., 25% of the patients had recurrence while the outcome was excellent in the cases where the cysts had been surgically removed without rupture. This highlights the need for long-term follow-up using clinical and radiological methods (CT scan, conventional MRI scan, and MR spectrometry) [12]. Medical treatment with albendazole seems to be beneficial both preoperatively and postoperatively. It is recommended that for treating brain hydatid cyst, the size of the cyst, multiplicity, location and neurological deficit must all be taken into consideration [13].

CONCLUSION

In children, intracranial hydatid cysts can grow to enormous sizes and the cases can remain neurologically

intact, rarely requiring emergency surgery. For this reason, severe headache in childhood should be taken into consideration carefully in countries where hydatid disease is seen. In case of emergencies, such as midbrain herniation with cystic lesions identified by CT scan, the possibility of hydatid cysts should be considered where the infection is endemic and the team should be prepared to remove the cyst without rupture. Our patient, who received emergency surgery, was discharged after one week following optimal neurological exams.

Author Contributions

Touzani Soumaya – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Bechri Brahim – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Joulali Toufik – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Berdai Mohamed Adnane – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Labib Smael – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Harandou Mustapha – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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