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Primary hydatidosis of the central nervous system: A retrospective study of 39 Tunisian cases

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ABSTRACT

Objective: To analyze epidemiological characteristics, clinical symptoms, radiological aspects, treatment and outcome of central nervous system hydatidosis and compare our results with those reported in literature.
Patients and methods: In our retrospective study, we reviewed 39 cases of primary central nervous system hydatid cysts operated on in our hospital between 1998 and 2007.
Results: There were 20 male and 19 female patients (sex-ratio M/F = 1.05) between 2 and 68 years of age (mean = 26.5 years). Thirteen of the patients were children (33.3%) with a mean age of 6.8 years and 26 were adults (66.7%) with a mean age of 36.3 years. The location of hydatid cysts was intracranial in 27 cases (69.2%) and spinal in 12 cases (30.8%). Headache and motor deficits were the predominant symptoms in patients with intracranial hydatidosis whereas back pain and spinal cord compression syndrome were the most frequent clinical presentations in patients with spinal hydatidosis. All patients underwent surgical resection of the cyst. Pathologic findings were consistent with hydatid cyst in all cases. During the follow-up period which ranged between 12 months and 5 years, 12 patients had recurrence (30.7%). Only one patient with intracranial hydatid cyst died postoperatively due to anaphylactic shock.
Conclusion: Despite all the advances in imaging techniques and therapeutic methods, central nervous system hydatidosis remains difficult to cure and patient outcomes are not satisfactory especially in case of spinal involvement due to the high incidence of recurrence.

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1. Introduction

Despite all the advances in imaging techniques and therapeutic methods, central nervous system (CNS) hydatidosis remains a serious health problem in Tunisia that is associated with high morbidity and mortality rates. Even though several reports of CNS hydatid cysts have been published in literature to date, only a small series focusing on both spinal and cerebral involvement is encountered [1]. In this paper, we report our experience with central nervous system hydatidosis over the past 10 years. Our aim was to analyze epidemiological characteristics, clinical symptoms, radiological features, treatment and outcomes of 39 patients who were surgically treated at our institution. Our results are analyzed in comparison to a review of the literature.

2. Patients and methods

We undertook a retrospective study of 39 patients who were operated on for central nervous system hydatid cysts at the neurosurgery department of the national institute of neurology of Tunis between January 1998 and December 2007. The cases were retrieved from the files of the Neurosurgery registry of the same hospital. Clinical records and microscopic slides of each patient were available for review in all cases. Clinical data, radiological investigations, treatment and outcome were retrospectively analyzed. Eighty percent of the patients came from rural areas. All patients underwent neurological and neuroimaging evaluation during the preoperative period. All specimens were surgically obtained. Tissues were fixed in 10% phosphate buffered formaldehyde, embedded in paraffin and sections were prepared for routine light microscopy after staining with hematoxylin and eosin. Abdominal ultrasonography and chest radiographs were performed after the diagnosis of hydatid disease during either the preoperative or the postoperative period. Patients with another visceral localization were excluded from this study. Patient confidentiality was maintained.

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Table 1
Distribution of 39 cases of central nervous system hydatid cysts by patient age.

	Children	Adults	Total
Intracranial hydatid cysts	12 (44.4%)	15 (55.6%)	27 (69.2%)
Spinal hydatid cysts	1 (9.4%)	11 (91.6%)	12 (30.8%)
Total	13 (33.3%)	26 (66.7%)	39 (100%)

3. Results

3.1. Clinical findings (Table 1)

Our study comprised 20 male and 19 female patients (sex-ratio M/F = 1.05) between 2 and 68 years of age (mean = 26.5 years). There were 13 children (33.3%) between 2 and 12 years of age (mean = 6.8 years) and 26 adults (66.7%) between 18 and 68 years of age (mean = 36.3 years).

3.1.1. Intracranial hydatid cysts

There were 14 male and 13 female patients (sex-ratio M/F = 1.07) with intracranial hydatid cysts ranging in age between 2 and 68 years (mean = 25 years). Twelve patients were children (44.4%) between 2 and 12 years of age (mean = 6.4 years) and 15 were adults (55.6%) between 18 and 68 years of age (mean = 39.8 years). The delay from onset of symptoms to diagnosis ranged between 1 week and 1 year. The presenting clinical symptoms were dominated by headache in 21 cases (77.7%), motor deficits in 11 cases (40.7%) and vomiting in 9 cases (33.3%) while visual disturbances and seizures were present in 6 cases (22.2%) and 5 cases (18.5%), respectively. Papilloedema was seen in 12 cases (41.4%) and positive Babinski sign was present in 5 cases (18.5%). Other symptoms included mental changes with irritability ($n = 3$), slackening of work ($n = 2$) and psychotic syndromes ($n = 2$).

3.1.2. Spinal hydatid cysts

There were six male and six female patients with spinal hydatid cysts ranging in age between 12 and 48 years (mean = 30 years). Eleven cases were adults (91.6%) between 24 and 48 years of age (mean = 31.5 years) and only one case was a child of 12 years old. The delay from onset of symptoms to diagnosis ranged between 2 weeks and 1 year. Most of the patients presented with back pain (83.3%) followed by sensory disturbance in seven cases (58.3%), bowel or bladder dysfunction in six cases (50%), paraplegia in five patients (41.6%), weakness of limbs in one case (8.3%) and radicular pain in one patient (8.3%).

3.2. Radiological findings and localization of hydatid cysts

Diagnostic imaging techniques included CT scan in all cases and MRI in 15 cases. Preoperative diagnosis was accurately made in 36/39 cases (92.3%). Two cases of spinal hydatidosis were misdiagnosed preoperatively as having disc herniation and spinal tumor and one case of cerebellar hydatidosis was misdiagnosed preoperatively as an arachnoid cyst. The cysts were supratentorial in 26 cases (66.7%) and infratentorial in 13 cases (33.3%).

3.2.1. Intracranial hydatidosis

Intracranial hydatid cysts presented as spherical, cerebrospinal fluid isodense lesions with no or minimal rim enhancement on CT scans (Figs. 1–4). Intracranial hydatid cysts ranged in size from 3.4 to 16 cm (mean = 8 cm) and produced considerable mass effect in the form of compression of the ipsilateral ventricles and shift of the midline structures. The sites of the cerebral cysts were as follows (Table 2): one lobe affected ($n = 14$), two lobes affected ($n = 9$) and three lobes were affected in three cases. Fifteen cysts were located in the left cerebral hemisphere and 11 cysts were located in the right one. One patient had a solitary cyst in the right hemisphere of the cerebellum. The hydatid cyst was solitary in 26 patients (96.3%) and multiple in only one case.

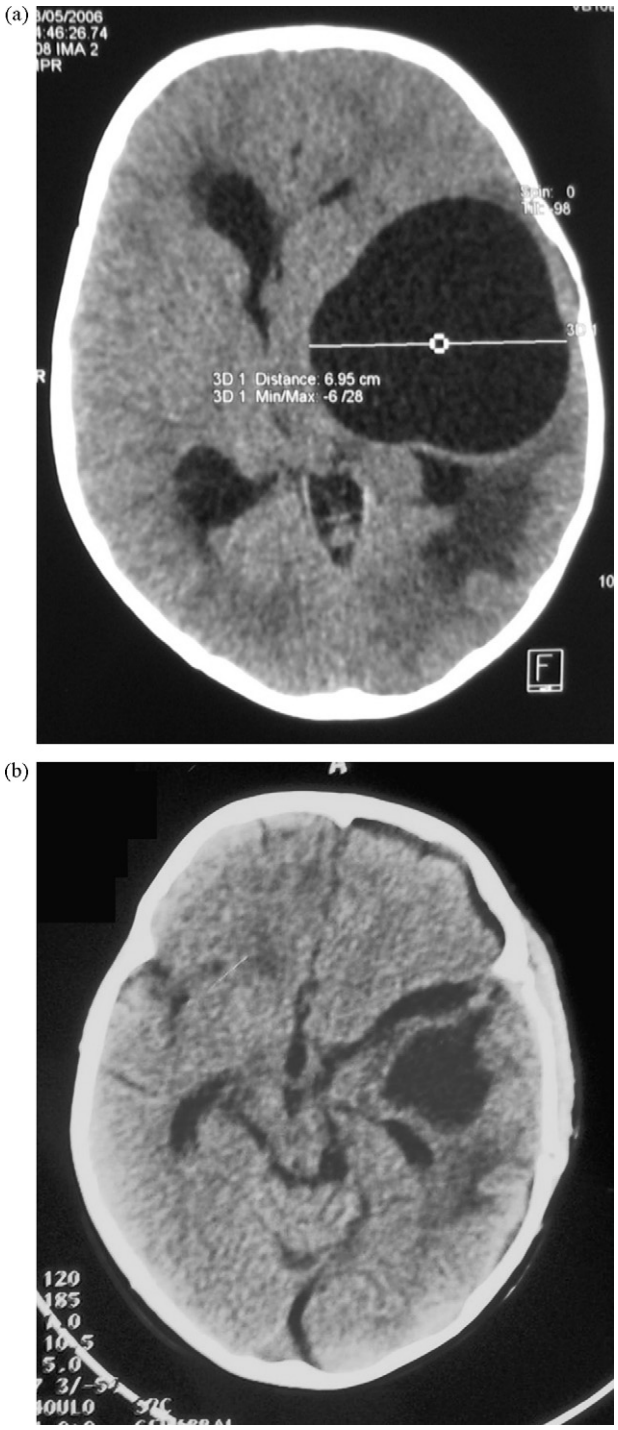

Fig. 1. (a) Preoperative CT scan revealing a well-circumscribed spherical hypodense cystic lesion of the left parietal lobe measuring 6.95 cm with midline shift. (b) Postoperative CT scan of the same patient showing a hypodense sequellar area within the same site of the excised cyst.

3.2.2. Spinal hydatidosis (Table 2 and Figs. 5 and 6)

Spinal echinococcosis involved the thoracic portion of the spine in seven cases (58.3%), the thoraco-lumbar region in two cases (16.7%), the lumbar region in one case (8.3%), the sacral and lumbar region in one case (8.3%) and the cervico-thoracic region in one case

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