Primary intracranial and spinal hydatidosis: a retrospective study of 21 cases

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Objective: To analyse the epidemiological characteristics, clinical symptoms, radiological aspects, treatments, and outcomes of primary central nervous system (CNS) hydatidosis and compare our results with those observed for secondary intracranial hydatidosis.

Patients and Methods: We retrospectively reviewed 21 cases of primary CNS hydatid cysts operated on at the First Affiliated Hospital of Xinjiang Medical University between 1996 and 2010.

Results: Of the 21 primary cases, the vast majority were intracranial hydatidosis patients (20 cases, 95.24%). Only one patient had spinal hydatidosis. Unlike previously published reports, we found that intracranial hydatid cysts were more common in adults (80.96%) than in children (19.04%), with a slight male predominance (M/F = 1.1). All symptoms, including vomiting, nausea, and focal neurological signs, resulted from the increased intracranial pressure, which was closely associated with the cyst location. For the spinal hydatidosis patient, the primary symptom of back pain was indicative of spinal cord compression syndrome. All cysts in the 21 primary cases were pathologically similar. The recurrence percentage was 28% over 12 years. Two patients with multiple intracranial hydatid cysts died due to foramen magnum herniation.

Conclusion: Despite imaging and therapeutic advances, CNS hydatidosis remains difficult to treat, and severe complications and the high incidence of recurrence result in unsatisfactory outcomes.

Keywords: Intracranial, Spinal, Hydatidosis

Introduction

Human echinococcosis is caused by infection with larval forms (metacestodes) of *Echinococcus* (*E.*) tapeworms found in the small intestine of carnivores, which is considered as one of the most geographically widespread zoonoses in the world. It remains highly endemic in pastoral communities, particularly in the regions of Middle Eastern Arab countries, China, India, Eastern Europe, Australia, New Zealand, and Latin American countries, but rare in Western Europe, North America, and USA.¹⁻⁴ Xinjiang, where probably over 50% of CE cases have been clinically reported, is considered to be the largest endemic area of echinococcosis in China.⁵⁻⁸ A large amount of clinical data was summarized by Xu⁸ from

58 hospitals in Xinjiang as follows: In 21 560 CE cases, 10 706 were male (49.7%) and 10 854 were female (50.3%), and the number of these cases under 3 years of age was 6209 (28.8%). In 45 (0.2%) cases the cysts were located in the brain. In this highly endemic area, the domestic dog is the main definitive host of *Echinococcus granulosus*, with sheep acting as the main intermediate host. There are an estimated 23 million sheep and two million dogs in the Xinjiang region. Direct contact of children or adults with dogs and ingestion of water, vegetables, or food contaminated by *Echinococcus* eggs are probably important in transmission of this disease.⁹ Under the World Health Organization's classification of hyperendemicity for *E. granulosus*,¹⁰ i.e., an annual surgical incidence of greater than 10 per 100 000 population and prevalence rates greater than 50% in sheep, virtually the whole of west, northwest, and parts of central China can be included. Data from surgical records in XUAR indicated that the highest incidence (>80 per 100 000 per annum) was observed in Han Chinese, especially from settled rural agricultural

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communities.¹¹ However, active mass ultrasound screening surveys, performed in traditional semi-nomadic pastoral communities of Kazakh or Mongolian ethnicity, revealed a higher incidence of CE infection.^{12–14} Primary hydatid cysts of the brain are rare and account for approximately 5% of all echinococcosis infections.¹⁵ Despite advances in imaging techniques and therapeutic methods, intracranial hydatidosis remains a serious health problem in Xinjiang and is associated with high morbidity and mortality rates. Here, we report our experience with primary central nervous system (CNS) hydatidosis over a 15-year period (1996–2010). We analysed epidemiological characteristics, clinical symptoms, radiological features, treatments, and outcomes of 21 patients who underwent surgery at our institution.

Patients and Methods

We reviewed the files of 21 patients who were operated on for primary CNS hydatid cysts at the First Affiliated Hospital of Xinjiang Medical University between January 1996 and December 2010. The cases were retrieved from the operation registry books of the Neurosurgery department. Clinical data, radiological investigations, treatments, and outcomes were retrospectively analysed. All of them underwent preoperative neurological and neuroimaging evaluations, and surgical specimens were obtained for each case. Tissues were fixed in 10% phosphate-buffered formaldehyde, embedded in paraffin, sectioned, and stained with haematoxylin and eosin for routine light microscopy assessment. Preoperative abdominal ultrasonography and chest radiographs were performed, and patients with visceral localization were excluded from the study.

Results

Intracranial echinococcosis

Clinical findings

Our study included 11 male and 10 female patients (M/F = 1.1) between 4 and 61 years of age (median = 26.5 years). Ninety per cent of the patients came from rural areas. There were four children (19.04%) between 4 and 12 years of age (median = 5 years) and 17 adults (80.96%) between 18 and 61 years of age (median = 36 years). There were one spinal and 20 intracranial hydatidosis cyst patients. The cyst was located in the supratentorial area in 13 patients, and seven patients had cerebellar cysts. The interval from symptom onset to diagnosis ranged between 10 days and 2 years. The presenting clinical symptoms were headache in 19 cases (90.5%), motor deficits in 12 cases (57.1%), and vomiting in 10 cases (47.6%), and visual disturbances and seizures were present in seven (33.3%) and six cases (28.6%), respectively. In two patients, coma was the first clinical manifestation. Papilloedema was observed in 11 cases (52.4%), and a

Figure 1 *Echinococcus granulosus* radiological findings. (A) Preoperative computed tomography (CT); (B) postoperative CT; (C) T1-weighted postoperative magnetic resonance imaging (MRI); (D) T2-weighted postoperative MRI.

positive Babinski sign was present in six cases (28.6%). Other symptoms included mental changes with irritability ($n = 4$), decreased productivity ($n = 1$), and psychotic symptoms ($n = 3$) (see Table 1).

Radiological findings and hydatid cyst localization

All cases underwent plain X-ray, computed tomography (CT) scanning, and magnetic resonance imaging (MRI). Preoperative diagnoses were accurately made in 19/21 cases (90.5%). Two cases were preoperatively misdiagnosed as tumours. The cysts were supratentorial in 13 cases (61.9%) and infratentorial in seven cases (33.3%). Intracranial hydatid cysts were spherical cerebrospinal fluid-isodense lesions with no or minimal rim enhancement on CT scans (Fig 1). Intracranial hydatid cysts ranged in size from 4.5 to 17 cm (mean = 7 cm) and produced a considerable mass effect in the form of ipsilateral ventricle compression and midline shift. Cerebral cyst involvement was as follows: one lobe affected ($n = 13$), two lobes affected ($n = 6$), three lobes affected ($n = 2$). The left and right cerebral

Table 1 Clinical manifestation of patients with primary intracranial hydatid cyst

	Number of cases (percent %)
Headache	19 (90.5)
Motor deficits	12 (57.1)
Vomiting	10 (47.6)
Visual disturbance	7 (33.3)
seizures	6 (28.6)
Coma	2 (9.5)
Papilloedema	11 (52.4)
Mental changes	8 (38.1)

Figure 2 *Echinococcus multilocularis* radiological findings. (A) T1-weighted postoperative magnetic resonance imaging (MRI); (B) T2-weighted MRI; (C) enhancement MRI.

hemispheres contained 15 and six cysts, respectively. One patient had a single cyst in the right cerebellum (Fig 2), and one had a hydatid cyst in a thoracic and sacral segment with vertebral body involvement.

Treatment

All patients underwent surgical cyst excision and received postoperative antihelminthic agents. Dowling's technique was used to execute the cyst totally by hydrostatic expulsion, that is, by forcing saline solution around and beneath the cyst. If the cyst ruptured inadvertently during surgical manipulation, the cyst was punctured, its contents were aspirated, and the shrunken cyst wall was extirpated. This method is known as PAIR. Accidental intra-operative cyst rupture occurred in six patients (28%). Two of our patients, including the one who initially presented with coma, died in the early postoperative period due to herniation. Other postoperative complications included seizures ($n = 1$) and hydrocephalus ($n = 1$).

Follow-up

The follow-up period ranged from 13 months to 15 years, and neurological status improved in all patients. However, recurrence and residual disease were observed over time. Six patients (28%) had recurrence, and four experienced multiple recurrences. The average time to recurrence was approximately 1 year. Postoperative sequelae included slight hemiparesis ($n = 5$) and cognitive dysfunction ($n = 3$).

Spinal echinococcosis

During the past 15 years, we diagnosed and treated only one patient with primary spinal hydatid disease. He presented with persistent back pain that was not responsive to usual conservative treatment, gradually increasing neurologic deficit of the lower limbs, and sphincter muscle dysfunction. Plain X-ray showed multiple, well-defined, osteolytic expansile cavitary areas at L2-S1 without periosteal reaction or sclerosis. Ultrasonography of the abdomen did not reveal any abdominal organ involvement at the time of first presentation. Computed tomography scan showed multiple cystic, osteolytic expansile lesions at L2-S1 vertebral bodies without enhancement of the lesion or the margins on the intravenous (IV) contrast study.

Magnetic resonance imaging showed multiple cystic fluid-filled lesions with thin walls and irregular branching resembling a grape bunch at L2-S1 levels on the axial, sagittal, and coronal images of the spine. The patient underwent surgery to excise the cysts and had a laminectomy performed through the posterior approach for neurologic decompression at the level of spinal involvement. At the time of surgery, multiple pearly, shiny, grape-like cysts were seen bulging from the spinal canal with multi-level paraspinal muscle involvement, which required extensive decompression and debridement through wide laminectomy. After the surgery, the patient received Albendazole (ABZ) for 1 year. At the last follow-up, the cysts had not recurred.

Discussion

A series with at least 20 total cases comparing child and adult patients revealed a range of 60–93% of cases occurring in children, with male predominance.¹⁶ Spinal involvement is also uncommon and is found in less than 1% of all cases.¹⁷ In our series, intracranial hydatid cysts were more common in adults (80.96%) than in children (19.04%), but we also observed a slight male predominance (sex ratio M/F = 1.1). The hallmark of this infection is the development of hydatid cysts in various organs, most commonly the lungs and liver. Cerebral manifestation of the disease is rare, occurring in about 2–3% of all *E. granulosus* infections.^{18,19} Cerebral hydatid cysts are oftensupratentorially localized in the terminal branches of the middle cerebral artery, usually temporo-parieto-occipital.^{20,21} Spinal echinococcosis is usually extradural.²² In our series, the cysts were supratentorial in 13 cases (61.9%) and infratentorial in seven cases (33.3%), only one patient had hydatid cysts with vertebral involvement. Clinical features of cerebral hydatid cysts are closely related to their localization. The major symptoms are headache, vomiting, nausea, and focal neurological signs, which all result from increased intracranial pressure. In our series, the presenting clinical symptoms in patients with intracranial hydatidosis were headache in 19 cases (90.5%), motor deficits in 12 cases (57.1%), and vomiting in 10 cases (47.6%). The patient with spinal hydatidosis presented with back pain, sensory disturbance, and bowel/bladder dysfunction.

CT scanning revealed solitary, homogeneous, spherical, and large parenchymal cysts with well-defined borders and without perifocal oedema. Cyst fluid density is the same as that of cerebrospinal fluid.²³ MRI is the preferred imaging modality when spinal hydatid disease is suspected and characteristically shows a lesion resembling a bunch of grapes. This unique feature can help in distinguishing hydatid infestation from spinal tuberculosis. The cyst walls are

thin and regular without septations. The presence of a markedly hypointense cyst wall on T1- and T2-weighted images and the absence of wall enhancement with gadolinium are also characteristic of hydatid disease. Although radiological features are very characteristic, definitive diagnosis of hydatid disease is based on histopathological examination of a surgical specimen. Even with the advances in CT and MR imaging, certain diagnostic problems remain. Some of these are related to changes in cyst structure, which can generate CT scans that differ considerably from classic images. These include infectious changes in the cyst and dead cysts. In cases that involve multiple lesions, smaller cysts located in and among larger ones may go undetected, even on CT and MR images. In our series, all cases underwent CT scanning and MRI. Preoperative diagnoses were accurately made in 19/21 cases (90.5%). Two cases were preoperatively misdiagnosed as tumours.

Total surgical removal of CNS hydatid cysts remains the treatment of choice. Many research studies suggest that the Dowling technique is the most effective and widely used method.^{24,25} It employs the use of hydrostatic assistance to dissect the fragile cyst wall from the brain parenchyma.²⁶ To avoid cyst rupture, the following criteria are essential during the procedure: creation of a large flap; adequate cortical opening to expose most of the cyst; meticulous cortical dissection; maintaining a right plane between the cyst and parenchyma; proper patient positioning; and the Valsalva manoeuvre.^{24,25} The technique is also suitable for cysts located deep or near the ventricular wall. To evacuate these cysts totally cortical opening and virtual structure retracting are inevitable. It is necessary to adopt microneurosurgery techniques; meticulous brain dissection; and adequate exposure during the procedure. In the case of multiple cerebral hydatid cysts, the largest one must be targeted first and then the other cysts must be removed during the same session.²⁴ The successful removal rate of intact intracranial hydatid cysts is approximately 60–70%.²¹ However, another long-term follow-up study confirmed that the outcomes of intracranial hydatidosis surgery without intra-operative cyst rupture are excellent.²² Nur Altinörs *et al.*¹ reported that out of 330 patients with hydatidosis who underwent operation, 207 cysts were executed successfully. In 86 cases (25.6%), the cysts ruptured inadvertently during surgical manipulation. In 35 cases (10.42%), the cyst was punctured, its contents were aspirated, the cystic cavity was irrigated with a hypertonic solution, and the cyst shrunk. This method is known as PAIR. Recurrence was reported in 56 patients (16.67%) at different periods postoperatively. A variety of post-operative complications were encountered in 32 patients (9.52%). There were 34 deaths in patients

with intracranial hydatid cysts, accounting for an overall mortality rate of 10.12% (34 of 336). The surgical mortality rate was 8.48% (28 of 330). In our series, intra-operative cyst rupture occurred in six intracranial cases (28%), which was slightly higher compared to other studies in the literature. Surgery may cure the patient completely, but sometimes it has certain risks, including anaphylactic reactions, secondary echinococcosis, and transient or permanent neurofunction deficit. Pre- and postoperative administration of antiparasitic drugs may be considered to sterilize the cyst, decrease the chance of anaphylaxis, decrease the tension in the cyst wall, and reduce the recurrence rate.^{25,27} But medical treatment is not the alternative therapy, it is only indicated for inoperable patients with echinococcosis of CNS. Albendazole is currently the drug of choice. It is less limited by the patient's status than surgery. Standard dosage-ABZ for 3–6 months produces an average of 30% cure.²⁸ Adverse effects of ABZ include hepatotoxicity, severe leucopenia, thrombocytopenia, alopecia,²⁹ embryotoxicity, and teratogenicity.³⁰

In our series, intra-operative cyst rupture occurred in six intracranial cases (28%), which was slightly higher compared to other studies in the literature. Medical treatment with ABZ in cases of recurrences, intra-operative cyst rupture, and giant or multiple hydatid cysts seems to be beneficial both pre- and postoperatively. A long-term follow-up study confirmed that the outcomes for intracranial hydatidosis surgery without intra-operative cyst rupture are excellent.²²

The spinal cyst in the study had not recurred at the time of last follow-up. Hydatid disease usually affects the soft tissues first and bones are involved later. The neurologic complications are the result of invasive intradural and extradural growth of the cysts causing direct compression. Destruction of the bones causes mechanical instability and secondary neurologic damage. This mixed result poses a confusing picture for diagnosis and treatment planning.^{31–34} MRI is superior to CT for demonstrating neural involvement.³⁵ Usually, posterior spinal decompression through laminectomy and debridement of paravertebral lesions is the initial surgery.^{31,36} The aim of surgery is the removal of all of the cysts early in the course of the disease.³¹ In a multi-centric study of a large series in Turkey, Nur Altinörs *et al.*¹ reported intra-operative rupture of spinal cysts in 44.4% of cases. Efficacy of ABZ for primary bony hydatid involvement is questionable; postoperative ABZ therapy seems to only retard recurrence.^{31,37} We attempted the intact removal of all of the visible cysts by wide laminectomy.

In summary, this retrospective study provides an overview of clinical symptoms, radiological features, treatments, and outcomes in 21 patients with CNS hydatidosis. In contrast to most studies reported in

the literature, most patients in this series were adults. Despite modern surgical and pharmacological therapies, hydatid disease of the CNS remains difficult to cure, and patient outcomes are less favourable due to the high incidence of recurrence.

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