Mixed cestode infection: an incidental association in an immunocompetent person

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SUMMARY

Echinococcus and *Taenia* infection are the two most relevant forms of cestode infection in humans. These infections occur worldwide, but with higher prevalence in developing countries like India, where poor hygiene facilitates their transmission. We report a case of a middle-aged man who presented with seizures and was found to have dual worm infection including *Taenia* and *Echinococcus*. The patient was treated with steroids and albendazole followed by PAIR (puncture, aspiration, injection of scolical agent and reaspiration). After 6 months of follow-up, the patient is asymptomatic and seizure-free without any relapse.

BACKGROUND

Human beings serve as both definite and intermediate hosts for cestodes. Clinical presentation of cestode infections varies from asymptomatic to severe life-threatening fatal disease. The therapeutic approach is often complex especially in mixed cestodal infection.

CASE PRESENTATION

A 42-year-old man presented with complaints of headache and two episodes of generalised tonic clonic seizure. The patient also gave history of vomiting, abdominal discomfort and heaviness on right side of the abdomen for last 6 months. There was no history of fever, cough, blurred or double vision. The patient was a strict vegetarian. There was no history of any addictions or any drug intake.

On examination, the patient was conscious and oriented with Mini Mental State Examination Score of 30/30. No signs of raised intracranial tension or meningeal irritation were present. The patient had hepatomegaly of 6 cm below the right mid-costal margin. Rest of the system examinations were normal.

INVESTIGATIONS

Haematological investigations revealed microcytic normochromic anaemia with haemoglobin of 7.6 gm/dl, total leucocyte count of 6200/mm³, differential count with 40% eosinophils, 50% neutrophils and 10% lymphocytes.

Liver function tests, renal function tests and serum electrolytes were within normal limits. Electrocardiography was also within normal limits. Mantoux test was negative.

Skiagram of the patient revealed multiple elongated calcifications (Rice-Grain calcifications) involving soft tissue of neck, chest, back and bilateral lower limbs, suggestive of cysticercosis (figure 1).

Ultrasonography of the abdomen revealed well-defined large cystic lesion with posterior acoustic enhancement in anterior segment of right lobe of liver (figure 2). Multiple internal septations and echogenic debris suggestive of hydatid sand were present. According to the WHO classification¹ it was categorised under Cystic Echinococcosis 3A (CE 3A) category, and according to Gharbis Classification,² it was type 3 cyst.

CT of head revealed multiple calcified lesions distributed diffusely in bilateral cerebral hemispheres without any perilesional oedema, suggestive of neurocysticercosis (figure 3).

ELISA for IgM of cysticercosis was positive.

ELISA for hydatid cyst was also positive.

Slit lamp examination of eye was normal.

Stool for ova and cyst was negative.

Serology for HIV was non-reactive.

TREATMENT

The patient was treated with steroid, albendazole (15 mg/kg/day in twice-divided doses) for 7 days along with anticonvulsant therapy. This was followed by PAIR (puncture, aspiration, injection of scolical agent and reaspiration) and again treatment with albendazole for 1 month.

OUTCOME AND FOLLOW-UP

After 6 months of treatment, the patient is asymptomatic and seizure free without any relapse.

DISCUSSION

Cestodes (commonly known as tapeworms) have different stages of their life cycle in humans. The most common infections by tapeworms include cysticercosis and echinococcosis. Cysticercosis is caused owing to infection by the larval stage of *Taenia solium* whereas echinococcosis is caused by *Echinococcus granulosus*. Exact incidence of both the infections is not known. Clinical features of cestode infection range from asymptomatic infection to severe life-threatening fatal disease.

Echinococcosis presents as hydatid cyst. Liver is the primary site, constituting nearly 70% of cases.³ It usually presents with upper abdominal discomfort and pain, poor appetite and abdominal lump. However, complicated cysts may manifest as jaundice, colicky pain, portal hypertension, ascites or compression of inferior vena cava.

In cysticercosis, the clinical manifestations are variable. Cysticercosis can be found anywhere in the body but is most commonly detected in the brain, cerebrospinal fluid, skeletal muscles, subcutaneous tissue or orbit. The clinical presentation of cysticercosis depends on the number, location and extent of associated inflammatory response.

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Figure 1 (A) Skiagram chest, (B) Skiagram right thigh, (C) Skiagram left thigh (D) Skiagram right leg (E) Skiagram left leg (F) Skiagram lumbar spine lateral view shows multiple elongated soft tissue calcifications with long axis along muscle fibres.

Neurological manifestations are most common which may include headache, seizures or features of increased intracranial tension. In studies from endemic areas, approximately 30% of cases of seizure disorder have evidence of neurocysticercosis on imaging studies.³

Imaging studies are the main diagnostic tests for Echinococcosis. Ultrasonography is the most common procedure

for diagnosing asymptomatic Echinococcosis. Ultrasonologically, echinococcal cyst can be classified using WHO Informal Group on Echinococcosis classification¹ or Hassen Gharbi classification² as shown in tables 1 and 2, respectively. Class CE 3b and above, according to WHO classification, represent inactive cysts.

Our patient had a well-defined cystic lesion with internal septations and echogenic debris in dependent portion (Hydatid

Figure 2 Ultrasonography of abdomen showing well-defined cystic lesion with internal septations and echogenic debris in dependent portion (Hydatid sand).

sand) suggestive of CE 3A category according to WHO classification and type 3 cyst according to Gharbi Classification.

Plain radiographs, CT and MRI are other imaging tests helpful in diagnosing Hydatid cyst.

Serological tests, ELISA, indirect haemagglutination (IHA) and immunoblotting also aid in diagnosis of Hydatid cyst. Eosinophilia is low grade or absent whereas routine laboratory investigations are non-specific. In cases with diagnostic dilemma, Fine needle aspiration cytology (FNAC) may be used to distinguish hydatid cyst from other cystic lesions.

Imaging studies (eg, CT and MRI scans) are the mainstay of diagnosis for neurocysticercosis. CT scans are more sensitive in identifying calcified lesions, which appear as 2–5 mm well-defined hyperdense nodules. Scolex inside a cystic lesion is diagnostic of cysticercosis. Antigen detection assays and enzyme-linked immunotransfer blot are serological tests helpful in diagnosis.

Figure 3 CT of head showing multiple diffusely scattered calcified granulomas without any perilesional oedema.

Table 1 The WHO Informal Group on Echinococcosis classification

Type of cyst	Status	Ultrasound features	Remarks
CL	Active	Signs no pathognomonic, unilocular and no cyst wall	Usually early stage, not fertile and differential diagnosis necessary
CE 1	Active	Cyst wall, hydatid sand	Usually fertile
CE 2	Active	Multivesicular, cyst wall and 'rosette-like'	Usually fertile
CE 3a	Transitional	Detachment of laminated membrane, 'water-lily sign,' less round-decreased intracystic pressure	Starting to degenerate
CE 3b	Transitional	Predominantly solid with daughter cysts	Starting to degenerate
CE 4	Inactive	Heterogeneous hypoechogenic or hyperechogenic degenerative contents, no daughter cysts	Usually no living protoscoleces, differential diagnosis necessary
CE 5	Inactive	Thick calcified wall, calcification partial to complete and not pathognomonic but highly suggestive of diagnosis	Usually no living protoscoleces

CE, cystic echinococcosis.

Ultrasonography and plain radiographs help in detection of cysticercosis in muscles.

The appropriate treatment depends on hydatid cyst characteristics (for hepatic cysts, size and stage are the most important criteria), the therapeutic resources available and the physician's preference. Surgery has been the traditional approach for the treatment of cystic echinococcosis (CE). However, chemotherapy, percutaneous procedures and a wait-and-watch approach has been successfully introduced now. These alternatives have replaced surgery as the treatment of choice in many cases.⁴

Albendazole should always be administered for at least 30 days after puncture. Antiparasitic drugs are playing an increasing role in the management of CE. Albendazole, 10–15 mg/kg/day, should be administered continuously without interruption. Albendazole usually causes solidification of small-sized and medium-sized CE1 and CE 3A cysts. In contrast, it is usually ineffective for larger (diameter more than 10 cm) CE1 and CE 3A cysts. Chemotherapy alone has no permanent effect on CE2 and CE 3b cysts.⁵

Optimal treatment of neurocysticercosis often includes symptomatic therapy for seizures and specific therapy for parasites (including antiparasitic drugs or surgical removal).^{6–8} Antiepileptic drugs are used to control the seizures.

Patients with calcified lesions do not have viable cysticerci and do not benefit from antiparasitic drugs.

In various randomised-controlled trials, it has been demonstrated that patients with parenchymal neurocysticercosis show

Table 2 Gharbi classification of hepatic echinococcal cysts

Type I	Pure fluid
Type II	Fluid collection, split-wall floating membrane
Type III	Fluid collection with septa, daughter cysts and honeycomb image
Type IV	Heterogeneous echographic pattern
Type V	Reflecting thick walls

more rapid resolution of parenchymal cystic lesions and have fewer generalised seizures when treated with corticosteroids and Albendazole (15 mg/kg/day in twice daily doses usually for 7–8 days) as compared with a placebo.^{9–11} Praziquantel (50–100 mg/kg/day in thrice daily doses) may also be used, but its benefits are not well defined. Patients with single enhancing lesion do well with or without antiparasitic therapy, but studies have demonstrated a slightly more rapid radiological resolution and fewer seizures when treated with steroids and/or antiparasitic drugs.

In our patient, the treatment was mainly targeted for echinococcus since the lesions of cysticercosis were disseminated and calcified. Our patient was treated with steroids, albendazole and PAIR of hydatid cyst.

Learning points

- ▶ *Echinococcus* and *Taenia* infection are the two most relevant forms of cestode infection in humans.
- ▶ Mixed cestode infection is a therapeutic challenge.
- ▶ Ultrasonography of cyst can determine the activity of echinococcal cyst.
- ▶ The appropriate treatment of cystode infection depends on patient characteristics, the therapeutic resources available and the physician's preference.

Competing interests None.

Patient consent Obtained.

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