Incidental finding of heart and brain echinococcosis in a patient with carbon monoxide poisoning

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Abstract: Heart and brain involvement of hydatid disease are rare findings in autopsy practice. We report the case of a 26-year-old man, admitted for a carbon monoxide poisoning, who died after 24 days of hospitalisation with hypoxic encephalopathy. After an underdiagnosed hospital ultrasound finding and a relevant history, the autopsy revealed not only the hepatic hydatid cyst but also an extensive microscopic echinococcosis that affected the heart and the brain. Those results, traumatic and non-traumatic can be difficult to interpret in relation to medico-legal investigation and patient history. Key Words: echinococcosis, hydatid cyst, carbon monoxide intoxication.

Hydatid disease can be caused in humans by Echinococcus granulosus – the most common, causing cystic echinococcosis; Echinococcus multilocularis, causing alveolar echinococcosis – rare but the most virulent; and Echinococcus vogeli (the rarest). The pathogen is endemic in southern Germany, Austria, Switzerland, and the Balkans.

Echinococcus granulosus, is a canine tapeworm, in which humans are intermediate hosts, as sheep, that ingest proglottids filled with eggs. The adult tapeworm is 3 to 6 mm, and consist of a scolex (the head), a neck, and a single proglottid with 100 - 1.500 eggs who lives inside the dog intestine (the definitive host). The intermediate host ingest a 30 - 35 microns egg that hatches and release oncospheres (larvae), which can disseminate through blood circulation in different organs, the most common being the liver (usually asymptomatic), but also, especially in younger patients, lungs, heart, brain, spine [1-3]. The cyst increase by about 1 cm per month, reaching 4 – 7 cm and containing an inner nucleated germinal layer with daughter cysts and protoscolecies, and an outer laminated anucleated layer. The cyst may release small amount of liquid, or burst with large leak, potentially causing anaphylaxis, superinfection, embolic spreading [4, 5].

CASE REPORT

A 26-year-old man, shepherd, without a medical history, was found in the morning (09:00) unconscious inside a transportation trailer that served as a shelter heated by a lit stove. It was taken by ambulance in a state of profound coma (GCS 3), presented a respiratory rate = 12/min, pulse rate =99/min, blood pressure = 130/70 mmHg, Sat02 = 85%, non-reactive pupils and myosis, and a diagnosis of carbon monoxide poisoning. Were performed tracheal intubation, mechanical ventilation, a NG probe, urinary probe, mechanical ventilation with balloon and oxygen 12 l/min.

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Later the patient was brought by helicopter and hospitalized for carbon monoxide poisoning and subsequent encephalopathy in the toxicology department. On admission at 13:55 p.m. the laboratory examinations showed acidosis (pH 7.18, HCO3- 13.4), hypoxemia (HbO2 94.4%), COHb moderately increased (2.3 %), hyperglycaemia (112 mg/dL), leucocytosis (16.500/ uL), liver cytolysis (AST = 68 U/L). Chest radiography revealed right interstitial opacities, tracheal secretion positive for Klebsiella and the abdominal ultrasound examination evidenced a simple serous cyst, 64 mm length in the fourth segment of the liver.

During hospitalization, the patient’s condition remained serious, with no improvements, 15 days after admission was performed a tracheostomy and after 24 days of hospitalization the patient died.

**MACROSCOPIC INVESTIGATION**

The autopsy performed 24 hours post mortem revealed signs of prolonged hospitalization and comatose posthypoxic encephalopathy: pressure sores, tracheostomy cerebral oedema, bronchopneumonia, pus-filled lung abscess (Figs 1 and 2), liver dystrophy but also a liver cyst with internal membrane and clear fluid (Figs 3 and 4). The primary cause of death was established as acute poisoning with carbon monoxide and consequent evolving post hypoxic encephalopathy and multiple organ dysfunction syndrome.

**Figure 1.** Macrosopy: An abscess below the posterior pleura of the left inferior lobe.

**Figure 2.** Macrosopy: Multiple left pulmonary abscesses.

**Figure 3.** Macrosopy: Hydatid cyst of the right hepatic lobe.

**Figure 4.** Macrosopy: Hydatid cyst of the right hepatic lobe - cut.
HISTOPATHOLOGY INVESTIGATION

Ethical approval
The case report has received ethical approval from the local ethics committee. Subsequently, tissue samples for microscopy analysis were taken after the informed consent, using a protocol approved by the local Bioethics Committee, in accordance to generally accepted international practice.

Tissue sampling and stains
Tissue specimens from brain, lung, heart, liver and kidney were taken for histopathology investigation. The selected tissue samples were fixed in 10% neutral buffered formalin (pH - 7) for 24–48 hours and paraffin embedded. Sections were cut at 5 μm and stained with standard HE, van Gieson and PAS.

RESULTS
On the examined sections of the brain, it was noticed numerous hooked protoscolices in the subarachnoidian space, accompanied by a heavily acute purulent inflammation (superimposed acute purulent meningitis - Fig. 5).

In the myocardium, one protoscolex was observed perifibrilar, in the interstitium and another one in the lumen of an isolated subepicardial coronary branch (Fig. 6).

The lung examination showed bronchopneumonia with diffuse alveolar damages and hyaline membranes. A large abscess was seen in the parenchyma, containing necrotic debris, as well as leucocytes and many bacterial colonies, surrounded by a thick pyogenic membrane (Fig. 7).

DISCUSSION AND CONCLUSION

Organ involvement
Diagnosis: The pathogen identification includes a hydatid wall of homogeneous eosinophilic cuticula, with

Figure 5. Microscopy (brain, HE): protoscolices in the subarachnoidian space with superimposed acute purulent meningitis.

Figure 6. Microscopy (heart, HE): a perifibrilar(UP) and an intracoronary (BOTTOM) protoscolex.

Figure 7. Microscopy (lung, HE): abscess with necrotic debris, leucocytes and many bacterial colonies, surrounded by a thick pyogenic membrane.
hooked protoscolices on the interior surface. Clinically the disease may present as echinococcosis of the liver (60% of cases), echinococcosis of the lung (20% of cases), and echinococcosis of the bone (2%). In some cases, the worm remains latent and suffers a spontaneous regression with calcification. These hydatids contain a sediment of dead protoscolices and granular calcification (hydatid sand).

**Cerebral involvement**

The involvement of Central Nervous System in hydatid diseases is reported to be 1-3% in literature. Brain involvement is considered a sign of the terminal phase of alveolar echinococcosis. In a recent paper was reported a 67-year-old female who had liver alveolar hydatid disease with brain and spinal intradural involvement [6]. Cerebral localization of E. granulosus especially affects children and is more frequently located in the supratentorial region. It can be life-threatening due to its localization in eloquent areas especially in the posterior fossa. Despite the benign nature of hydatid cyst, invasion of critical areas may cause significant mortality and morbidity in some patients. Urgent surgical decompression and adjuvant medical treatment must be employed as soon as possible in these patients. Supporting this idea, in another article is presented a clinical case of life-threatening brainstem compression in a child, due to a rare form of cystic echinococcosis, which was confirmed with biomolecular techniques [7]. Giant cerebral hydatid cysts were also reported in teenagers [8]. Even so, rare cases of cerebral echinococcosis are reported in the literature, one of them with particular intraventricular (left lateral ventricle) hydatid cyst in a 21-year-old adult female. In these cases, the possibility of infection with Echinococcus granulosus should be included in the differential diagnosis of raised intracranial hypertension in patients from endemic areas [9].

**Heart involvement**

The cardiac hydatid cysts are rare and may be found intracavitary or extracavitary. In a recent study involving 25 patients, the cardiac cysts were reported as intracavitary in 11 patients and extracavitary in 14 patients [10]. There is some controversy about how echinococcosis spreads to the heart (via haematogenous spread or direct extension from adjacent structures). According to the aforementioned study [10], it is thought that haematogenous spread is the main method of the distribution of cardiac echinococcosis, and the direct extension method from adjacent structures must be questioned. Other rare cases were also reported in the literature, with localization in the left atrium [11], right ventricle [12] and left ventricular wall [13].

Our case presented several specific features in terms of both evolution and legal medicine:

- Clinical examination revealed no signs of anaphylaxis, with a conclusive history of carbon monoxide poisoning.
- Primary hepatic hydatid cyst was incidentally diagnosed only at autopsy although there was a presumptive diagnosis by ultrasound in hospital.
- Intracerebral and intracardial protoscolices presence without membrane vesicles associated with meningitis was revealed only at microscopic examination. Correlating the aspects above it resulted that most likely the release of protoscolices occurred relatively recent, within a maximum of a few weeks, as a microscopic leakage from a primary source and prompted the death initiated by an acute carbon monoxide poisoning with secondary coma.

**Conflict of interest.** The authors declare that they have no conflict of interest concerning this article.

References