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# The first case of autochthonous human alveolar echinococcosis in the Slovak Republic (Case report)

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## Introduction

Human alveolar echinococcosis (AE) caused by the metacestode stage of *Echinococcus multilocularis*, is characterised by a tumour-like, infiltrative and destructive growth with the potential to induce a serious disease with a high mortality rate. In Europe the sylvatic cycle of *E. multilocularis* involves red foxes and rodents; domestic dogs and cats are also susceptible (as definitive hosts) and may become infected by predating wild small animals (intermediate hosts) (8). The mature segments (or eggs) of the "fox tapeworm" are excreted via the faeces to the environment. Eggs are very resistant and remain infective for several months (5), thus farmers, forest workers, tourists, forest fruits pickers, etc. are mainly exposed to the infection (5, 6).

Expansive spread of this cestode from endemic alpine regions to the Central Europe area has not stopped on the borders of our country. The first finding of *Echinococcus multilocularis* in red foxes in the Slovak Republic was recorded in 1999, primarily in the border regions (3), however, *E. multilocularis* infected foxes could be detected almost in the whole territory during the year 2000 (4). In this paper we present the first autochthonous case of alveolar echinococcosis in the Slovak Republic.

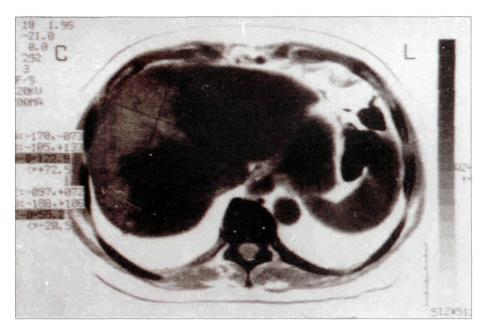


Fig. 1: Nodular hyperplasia of hepatic parenchyma with microcalcifications (arrow).

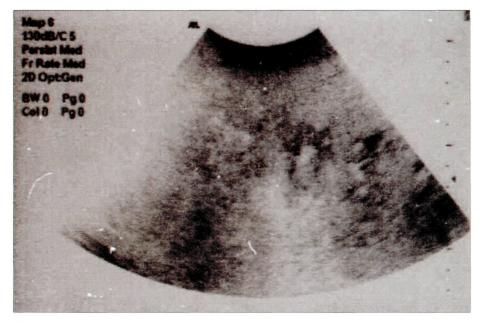


Fig. 2: Microcalcifications with central hypoechogen foci (arrows).

## Case report

The patient, a 69 years old woman living in a village in the northwestern region of the Slovak Republic, started to suffer from the dyspeptic problems accompanied by pain in the right epigastrium in 1994. She was treated symptomatically, the cause of her complaints remained unexplained. In September 1999 she was admitted to the hospital due to a painful biliary colic. Cholecystolithiasis was supposed due to abdominal ultrasound examination. The CT examination, however, showed nodular hyperplasia of the liver parenchyma with an irregularly shaped focus (10 cm in diameter) of hypoechogenic character with microcalcifications. Due to the suspicion of a possibly malignant liver tumour a biopsy of the focus was performed under ultrasound (US) control, a carcinoma, however, could be excluded histologically. Laboratory diagnostic examinations revealed an accelerated erythrocyte sedimentation rate and a slight increase of aminotransferases (GPT and GOT). The markers of viral hepatitis were negative and the level of bilirubin did not exceeded the standard level.

In November 1999 a surgical puncture of the focus was performed under CT controlling, yellow-green pus was obtained and the focus was drained. One week later a second unclearly encircled focus with irregular shadow-casting (microcalcifications) with central hypoechogenic foci in size 2 x 1 cm was found by ultrasound (US) examination (Figure 1, 2, 3).

Patient's serum was tested on specific (IgG) antibodies against *E. multilocularis* and *E. granulosus* antigens by ELISA and westernblot in the Parasitological Institute in Kosice and in the Department of Medical Parasitology of the Clinical Institute of Hygiene, University of Vienna. ELISA testing (with *E. granulosus* and *E. multilocularis* antigen, respectively) yielded positive test results in both institutes, western blot analysis (performed with different *E. multilocularis* antigen preparations in Kosice and Vienna, respectively) revealed the diagnosis: alveolar echinococcosis.

In January 2000 a second biopsy (during cholecystectomy) of the liver in the area of the described focus was performed. Histological findings gave evidence of alveolar echinococcosis (Fig. 4) and thus, confirmed the parasitological-serological diagnosis. The patient was admitted to the hospital and treated with mebendazol (Vermox®) in the dose of 1000 mg per day in the course of 3



Fig. 3: Echogenic area associated with cystic zones.

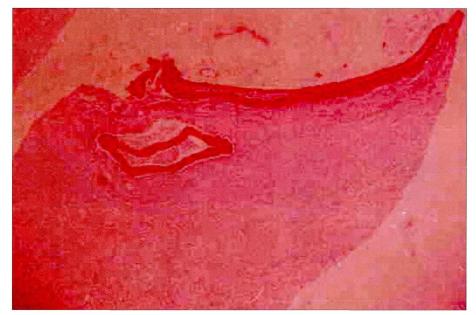


Fig. 4: Purplishly died PAS positive findings of alveolar echinococcosis.

weeks. Repeatedly, control US examinations after 6 weeks showed moderate regression. Subsequently the patient was treated with albendazol (Zentel®) (1000 mg/day; 4 weeks). Two months later the main lesion had decreased in size (5 cm in diameter), was of hyperechogenous character and showed no signs of reactivation.

The patient is still under ambulatory control and treatment, negative clinical side effects have not been recorded, so far.

### Discussion

Alveolar echinococcosis has been known since about 150 years when the pathologist Buhl (2) observed an "Alveolarkolloid" in the liver of a Bayuvarian patient for the first time. Fourty years later the Austrian physician Adolf Posselt diagnosed the first human AE cases in Austria; in addition he also collected all data of each AE case recorded worldwide (7). Due to Posselt's epidemiological records we know, that alveolar echinococcosis was prevalent in southern Germany, Switzerland and in Austria, but could not be observed in the eastern regions of Central Europe, at least until 1936, the year of Posselt's death.

The detection of *E. multilocularis* in Vulpes vulpes (red foxes) in nearly all regions of the Slovak Republic (3, 4), however, led to the expectation that alveolar echinococcosis might also occur in Slovakia. Due to the fact that alveolar echinococcosis and its nosology is rather limited among medical staff on one hand and that - on the other hand - the clinical symptoms of this helminthosis as well as the morphological structures of liver lesions (detectable by imaging techniques) might resemble other liver diseases (e. g. malignant neoplasm of the liver and the bile ducts, hypertrophic liver cirrhosis), also in our case - as in many other cases observed worldwide - the parasitic disease was diagnosed several years after onset of clinical symptoms. Our patient, a dog keeping and forest fruits picking up 69

years old female from the countryside of Northwestern Slovakia, however, must be infected many years (incubation period of alveolar echinococcosis: 5 - 15 years) before the appearance of clinical symptoms in 1994. The clinical picture of an acute cholecystolithiasis, finally, induced intensive radiological, histological and parasitological examinations. It has to be stressed that ELISA and western blot, performed in two different parasitological laboratories using different antigen prepa-

rations (Reiterova et. al., unpublished; 1) yielded an identical (species-specific) diagnosis. The sero-logical diagnosis could subsequently be confirmed by histological examination.

Based on the experiences with the therapeutic procedures in cystic echinococcosis, the patient received a mebendazole treatment for 3 weeks, thereafter albendazole (in 4 weeks courses) was administered to the patient. Surgical resection of the parasite was not carried out so far, particularly due to the advanced age of the patient and the obvious antihelminthics-induced regression of the liver lesions.

Due to the detection of the first autochthonous case of alveolar echinococcosis in the Slovak Republic medical staff should be trained to include this serious helminthic disease into differential diagnosis at patients suffering from symptoms deriving from the liver or the bile duct. Highly sensitive and specific serological tools are available in order to detect *E. multilocularis* infections of man in a quite early (and asymptomatic) stage.

#### Summary

We report the first case of autochthonous alveolar echinococcosis in a 69-years old woman in the Slovak Republic. The patient was admitted to the hospital due to a biliary colic. Ultrasound and Computed tomography examinations showed nodular hyperplasia of the hepatic parenchyma with an irregularly shaped focus of hypoechogenic character in size of 10 cm with microcalcifications. Haematological examinations were inconspicuous, biochemical tests revealed only slight increase of aminotransferases. The diagnosis of hepatic alveolar echinococcosis was based upon histopathological examination of bioptic samples, which were taken peroperationally during cholecystectomy and was confirmed by serological methods (ELISA, Western blot). The long-term therapy with benzimidazoles (ie. mebendazole and albendazole) stopped the growth of the parasitic liver lesions

Alveolar echinococcosis has to be included into differential diagnosis in Slovakian patients suffering from hepatic or biliary disorders.

#### Key words

Echinococcus multilocularis, alveolar echinococcosis, ELISA, western blot, Slovakia.

## Aknowledgements

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## Zusammenfassung

Der erste Fall einer autochthonen humanen alveolären Echinokokkose in der Slowakei (Fallbericht)

Wir berichten über den ersten autochthonen Fall alveolärer Echinokokkose in der Slowakischen Republik. Dabei handelt es sich um eine aus dem Nordwesten des Landes stammende 69jährige Frau, die mit Verdacht auf eine akute Cholezystolithiasis in stationäre Behandlung aufgenommen wurde, nachdem sie bereits 5 Jahre zuvor mit epigastrischen Schmerzen und anschließender symptomatischer Behandlung ärztliche Hilfe in Anspruch genommen hatte. Oberbauchsonographieund Computertomographieuntersuchungen erbrachten eine noduläre Hyperplasie des Leberparenchyms mit einem irregulär begrenzten, hyperechogenen Fokus (von 10 cm Durchmesser) mit Mikrokalzifikationen. Auffallende Laborbefunde waren eine Erhöhung der Blutsenkungsgeschwindigkeit sowie geringgradig erhöhte GOT- und GPT-Spiegel. Die Diagnose "alveoläre Echinokokkose" wurde aufgrund eindeutiger histologischer sowie parasitologisch-serlogischer Untersuchungen, die in zwei verschiedenen Instituten (Kosice und Wien) unter Verwendung verschiedener Antigenpräparationen durchgeführt wurden, gestellt. Die Patientin wurde vorerst mit Mebendazol behandelt, nach drei Wochen wurde auf Albendazol umgestellt. Das Präparat wurde bislang gut vertragen, die Leberläsionen haben sich unter der antihelminthischen Therapie deutlich verkleinert.

Die alveoläre Echinokokkose sollte bei slowakischen Patienten mit Beschwerden der Leber und der Gallenwege in die Differentialdiagnose einbezogen werden.

#### Schlüsselwörter

Echinococcus multilocularis, die alveoläre Echinokokkose, ELISA, Westernblot, Slowakei.

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