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Case Report

A Disseminated Echinococcosis Patient with Five Years Survival from Turkey: A Case Report

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Abstract
Echinococcosis is a parasitic disease characterized by cysts in especially liver and lung. We report a long-term survival of a 44-year-old female patient with disseminated echinococcal disease involving the brain, lung, liver, spleen, kidney, mediastinum, thyroid gland, parotid gland, pancreas, peritoneum, rectus muscle, pararenal area, left thigh, skin and breast tissue from Turkey in 2016.

Introduction

Echinococcosis is infection with larvae of the tapeworm *Echinococcus*. Four species of *Echinococcus* cause infection in humans. *E. granulosus* causing cystic echinococcosis and *E. multilocularis* causing alveolar echinococcosis (AE), are the most common species encountered in humans. The two other species, *E. vogeli* and *E. oligarthrus* are associated with "Neotropical echinococcosis" and less frequently associated with human infection (1).

Cystic echinococcosis is globally distributed and more frequently in rural areas. Alveolar echinococcosis is in the northern hemisphere, including central and northern Europe, Central Asia, northern Russia, northern Japan, north-

central United States, northwestern Alaska, and northwestern Canada (2). "In regions where cystic echinococcosis is endemic, incidence rates in humans can exceed 50 per 100 000 person-years; prevalence levels as high as 5–10% may occur in parts of Argentina, Central Asia, China, East Africa and Peru" (3). According to the studies performed in different regions and times in Turkey, the prevalence is 0.2-12% (all references in 4).

We report a long term survival of a patient with disseminated echinococcal disease involving the brain, lung, liver, spleen, kidney, mediastinum, thyroid gland, parotid gland, pancreas, peritoneum, rectus muscle, pararenal area, left thigh, skin and breast tissue from Turkey.

Case Presentation

A 44-year-old woman was admitted to Emergency Department of a hospital in Ankara which is the capital of Turkey because of fatigue, gait disturbance, dysphasia, dyspnea and weight loss in 2016. Medical history was notable that she had headache and personality changes for about ten years and was thought to have psychiatric disease by her households. She was housewife and lived in Ankara for 10 years. She lived in a rural area in northern Anatolia before Ankara. She had no known illnesses. She did not have any domestic animal but had a history of being in touch with cows and sheep in her childhood.

Her physical examination revealed that she was lethargic, disoriented and uncooperative, the temperature was 38.5 °C, the heart rate 121 per minute, the blood pressure 100/65 mmHg, the respiratory rate 30 breaths per minute, and the oxygen saturation 93% while the patient was breathing ambient air. Pupillary reaction to light is normal in both of eyes and she had anisocoria. Motor examination was normal for four extremities. She had no meningeal irritation signs. The lungs were clear on auscultation. The heart was tachycardic, with normal heart sounds.The remainder of the examination was normal. Her initial laboratory results were; C-reactive protein 1,8 mg/l (reference range (RR):0-0.8 mg/l),) total leukocyte count: 8600/uL (RR:4500-11000/uL), haemoglobin:11.9g/dl (RR:11.7-15.5), platelet: 509.000/mm^3 (RR :150000 450000/mm^3), liver and renal function tests were normal.

Computed tomography (CT) of the brain performed and revealed two cystic lesions and cranial magnetic resonance imaging (MRI) revealed two lesions, 36x33 mm in size in the left parietooccipital cortical-subcortical located, adjacent to the posterior interhemispheric fissure and 51x42 mm in size in the midline-left paramedian located in the posterior fossa,affecting both cerebellum (Fig.1).

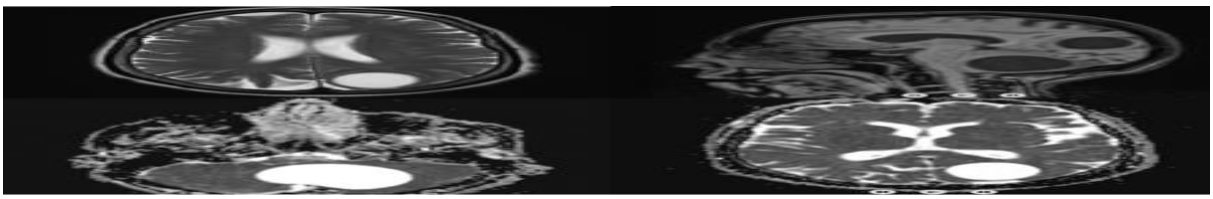

Fig. 1: Cystic lesions are observed in the cerebellar hemisphere and left parietal lobe in axial T2 and sagittal T1-weighted sequences in cranial magnetic resonance imaging. No significant diffusion restriction was observed in diffusion-weighted sequences

In thoracic CT, multiple cystic lesions, the largest of which was measured 3 cm in diameter, were observed in both lungs, and there were also multiple cystic lesions in the mediastinum and thyroid gland (Fig.2 and Fig.3).

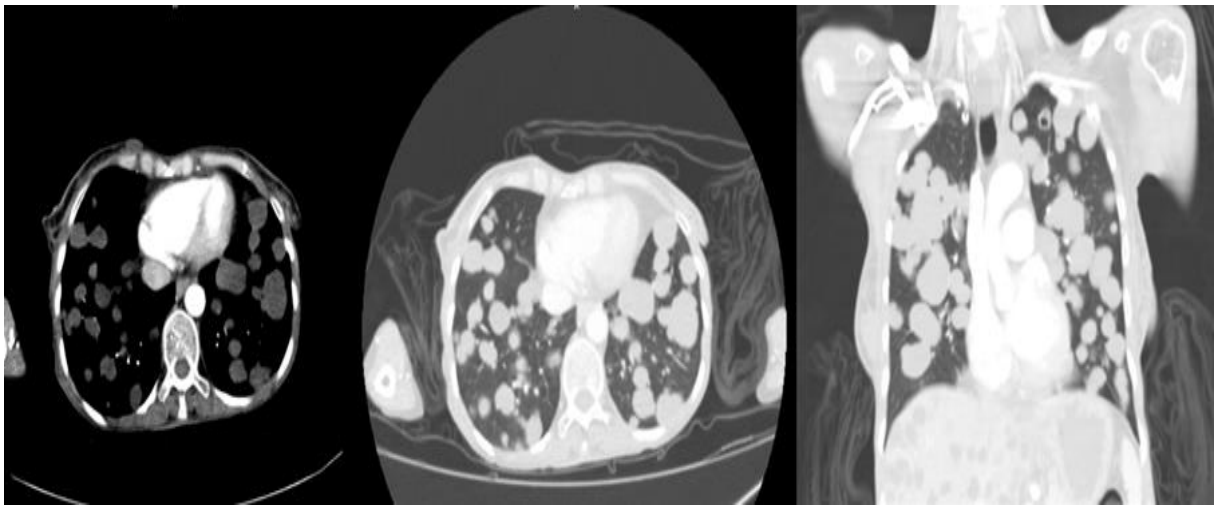

Fig. 2: Axial and coronal reconstruction view of contrast-enhanced CT scan show multiple cystic lesions in lungs

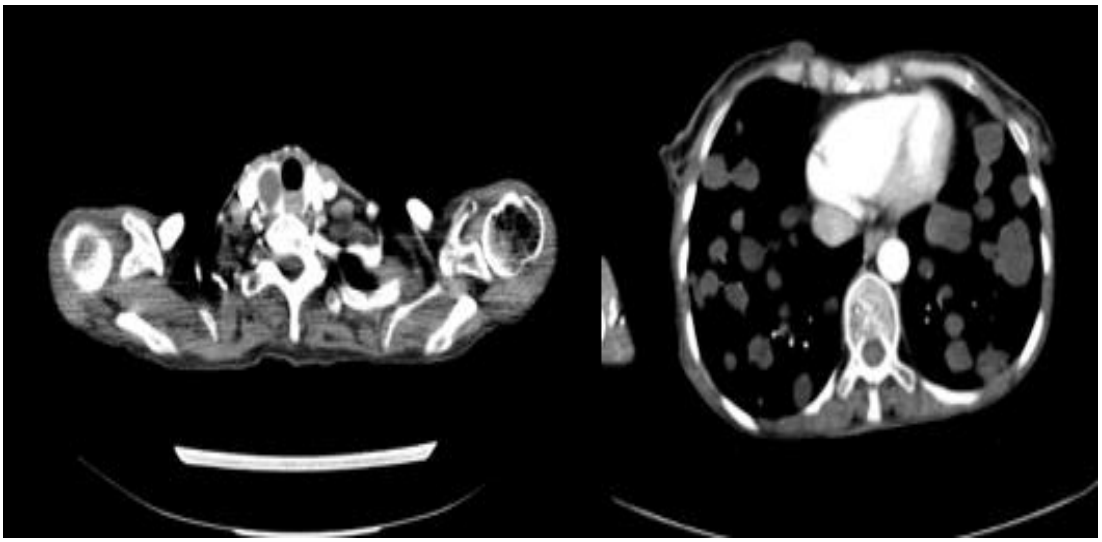

Fig. 3: Axial view of CT scan shows cystic lesions in the right thyroid lobe and subcutaneous cysts on the right side of anterior chest wall

Two pure anechoic cysts of 20 mm diameter in the right lobe and 18mm in the left lobe and another 18mm diameter cyst in the right paratracheal region were observed in thyroid ultrasonography. Abdominal CT with the administration of intravenous contrast material revealed multiple cystic lesions in liver, spleen, pancreas, peritoneum, obturator region, rectus muscle, kidney and pararenal region (Fig.4).

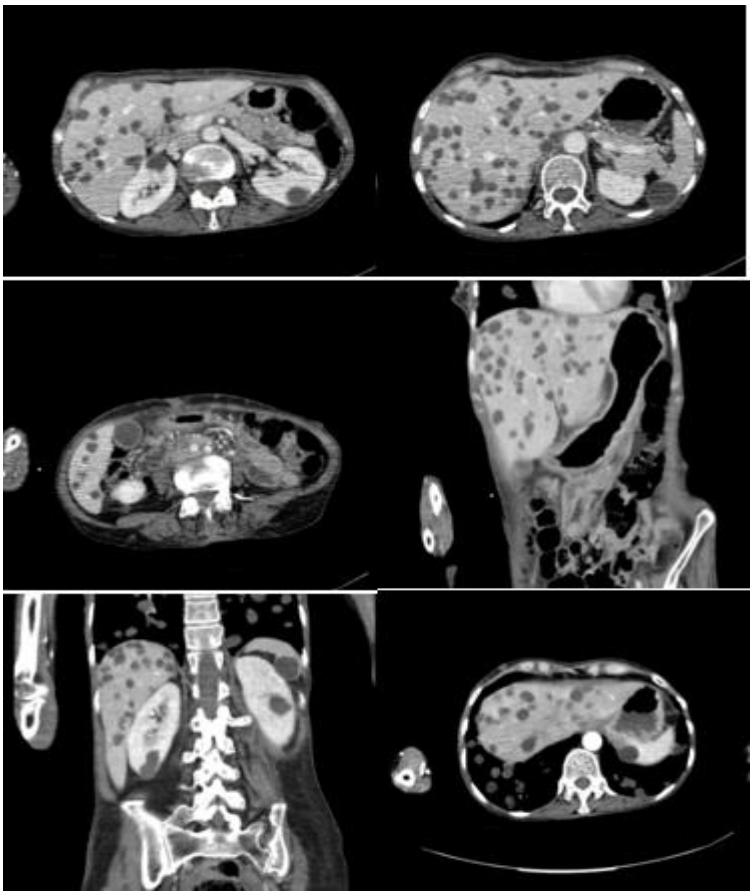

Fig. 4: Axial and coronal reconstruction view of contrast-enhanced abdominal CT shows multiple cystic lesions in liver, both kidneys, pancreas and spleen

Cervical and thoracic MRI revealed cystic lesions in parotid gland, thyroid gland, right parapharengeal fat tissue, right semispinalis capitis muscle, right side of trapezius muscle and T12 paravertebral muscle. She was admitted to the Neurosurgical Intensive Care Unit. Intracranial cysts excision was performed with an urgent operation on the admission day of patient. Indirect hemagglutination test was positive in titer 1/2560 and blood total IgE level was higher than 2000 IU/ml. Albendazole treatment was initiated. No intervention was planned for cysts in other tissues. The patient had an uncomplicated postoperative course and was discharged with oral albendazole treatment after one month of operation. The patient did not come to follow up within five years of her diagnosis. The phone number of the patient registered in the system was called and called for control in 2021. It was learned that the patient was alive and stayed in a nursing home. However, the details of her health status in 2023 could not be learned.

Discussion

We report a patient with disseminated echinococcal disease. Although more than 90% of cysts occur in the liver and lungs, there are case reports for almost all the organs involved with echinococcosis such as kidney, spleen, peritoneal cavity, prostat gland, skin and muscles, heart, brain, vertebral column (5, 6). An informed consent was obtained from her brother.

It is notable that these involvements were all together in our patient. Despite the ability of spread of echinococcal disease, dissemination of all the body is rarely reported. For multivisceral echinococcosis which is a condition that localization of cysts in more than one organ, several mechanisms are described. These mechanisms may lead to primary multivisceral cystic echinococcosis or to secondary (metastatic) cystic echinococcosis in which larval tissue spreads from the primary site and new cysts develop after spontaneous or trauma-induced cyst rupture or after release of viable parasite material during invasive treatment procedures (7, 8).

"Throughout the past 30 years, experimental studies probing the immunobiology of *E. granulosus* have begun to uncover an evolving story in which parasite immunomodulating proteins actively interact with innate and adaptive human immune processes to reduce the impact of a host response (9)". The factors that determine the final localizations of the metacestode of Echinococcus in a host are not clear but probably include anatomical and immunological characteristics of the host as well as the infection dose, parasite genotype. Risk factors for dissemination have not been yet understood since it is a rarely reported however; multiple cysts disseminated over several organs may be associated with delayed diagnosis and treatment.

Because cysts grow slowly, the host often tolerates it well and up to 60% of all cystic echinococcosis cases may be asymptomatic, although an unknown proportion may become symptomatic (5). Patients may become symptomatic with cysts in large diameter press to adjacent tissue or complications such as cyst rupture causing anaphylactoid reaction. This reported patient had headache and behavioral changes for several years depending on intracerebral cysts but had been thought to have a psychiatric illness. There are reported echinococcus cases with a misdiagnosis as behavioral change like our patient (10).

The mortality rate is estimated to be 0·2 per 100 000 population, with a case fatality rate of 2·2% which is higher in AE if not treated (5). Although disseminated involvement in our patient, her course of infection was not fatal.

Conclusion

This report indicates the importance of prevention and early diagnosis of echinoccoccus diseases especially in endemic regions or people with a history of travelling to endemic areas. In conclusion, a patient with a cystic lesion in any organ should be examined in terms of echinococcus and initiated appropriate treatment in especially endemic regions.

Acknowledgements

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Conflict of Interest

Non- declared.

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