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FIRST PORTABLE ULTRASOUND BASED SCREENING STUDY IN BULGARIA ON THE PREVALENCE OF CYSTIC ECHINOCOCCOSIS IN KARDZHALI DISTRICT

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ABSTRACT

PURPOSE: Cystic echinococcosis (CE) is a serious medical and social problem for the public healthcare in Bulgaria. The average incidence of the disease remains steady during the past 5 years-4.45/100000, while the average incidence rate for Kardzhali region was 8.02/100000. These data however do not reveal the morbidity of cystic echinoccoccosis in various settlements in the region. The aim of the present study was to investigate the prevalence and risk factors for CE using a portable ultrasound in Kardzhali District.

METHODS: From February till April 2014 abdominal ultrasound examination were performed in 642 volunteers including 302 (47%) males and 340 (53%) females, aged from 1 to 88 years. All underwent a questionnaire survey and abdominal ultrasound examination. The WHO-Informal Working Group on Echinococcosis standardized classification was used.

RESULTS: Volunteers from 4 villages in Kirkovo municipality, District Kardzhali, participated in the screening. Two patients with liver CE were detected, one of them relapse. The total prevalence of confirmed human CE (new and existing cases) was 1.25 (8/642) and ultrasound CE prevalence was 0.3% (2/642) in the current study.

CONLUSIONS: The CE prevalence of 0.3% indicates an important public health problem in rural areas of Kardzhali District demanding further portable ultrasound investigation.

Key words: Cystic echinococcosis, Ultrasound screening,

INTRODUCTION

Cystic echinococcosis (CE) is one of the world's most geographically widespread parasitic zoonoses (1). Domestic dogs are definitive hosts and livestock (sheep, goats, pigs, cattle, etc.) the main intermediate hosts. The disease is severely harmful to both humans and animals (2). In Europe the highest prevalence of cystic echinococcosis was found in the Mediterranean region (3, 4). Cystic echinococcosis is a serious medical and social problem for the public healthcare in Bulgaria (5, 6, 7, 8, 9). According to official data from National Centre of Infectious and Parasitic Diseases, the average incidence of the disease remains steady during the past 5 years-4.45/100000. In February 2014 a 22 year old man from Shoptsi, Kirkovo Municipality, Kardzhali District died from a severe anaphylactic shock after rupture of CE liver cyst. Because of that as well as for the fact that the average incidence rate of CE for Kardzhali District for the period 2008-2012 was 8.02/100000 (10,11) we decided to conduct ultrasound screening among this population.

Two specific CE mass screening surveys have been carried out in Bulgaria. Both studies were by serology (12, 13). The first was conducted in the 1974 -77 and seroprevalence was found as 6.21% among 6343 persons covered. In 104 persons (26.39%) the diagnosis was confirmed surgically (12). The second was carried out within the National Programme for Control of Echinococcosis in humans and animals conducted during 2004-2008. During the Programme implementation with ELISA and

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Western blot were tested 9390 sera samples from 9 districts of the country. Confirmed positive result was established in 107 sera samples and the seroprevalence was 0,68% (13). Kardzhali District was not included in these two studies.

The ultrasound (US) is the first choice among the various screening methods due to its easy and low cost applicability and high diagnostic success (14, 15). Ultrasound screening so far has not been performed in Bulgaria. The purpose of the study was to investigate the prevalence and risk factors for CE using a portable US in rural areas of Kardzhali District, where CE is known to be endemic.

MATERIALS AND METHODS

The pilot study was in Kirkovo municipality villages as a part of US screening survey for CE in Southeast Rhodope region. From February till April 2014 abdominal US examination were performed in 642 volunteers including 302 (47%) males and 340 (53%) females, aged from 1 to 88 years. By ethnicity 629 were Turkish and 14 Bulgarians. People were scanned by abdominal US operated by an experienced qualified gastroenterologist using a portable ultrasound scanner (FF – Sonic, UF – 3500, Fukuda Denshi Co., Ltd, Japan). The intra-abdominal organs were examined. Images with CE characters were graded according to WHO-Informal Working Group on Echinococcosis standardized classification into the following groups: undifferentiated cystic lesion (CL), active (CE1 and 2), transitional (CE3) and inactive (CE4 and 5) (16, 17). Detected cysts were measured and the CE positive cases were subjected for further investigation including serology and chest X-ray. A questionnaire with 22 questions included the information regarding volunteers demographic and social characteristics as well as their habits possibly related to CE were distributed to all participants for completion. For people who do not speak Bulgarian language questionnaires were completed with the help of translators. Univariate odds ratios were used to determine independent associations of risk factors in different variables including, gender, ethnic, dog owner, home slaughter, occupations, hygienic habits.

To summarize the results following methods were used - physical, imaging (ultrasonography, X-ray, CT), parasitological (serology - ELISA), epidemiological, sociological and statistical.

RESULTS

The area of study was in Kirkovo municipality, district Kardzhali located in the south Bulgaria. Kirkovo municipality is situated on 538 sq. km in the East Rhodopi Mountain and consists of 73 settlements, all of which are villages and hamlets. The average altitude is 523 m. According to the 2011census, it has a population of 21 916 comprising 3 main ethnic groups – Bulgarian, Turkish and Roma, as the dominant ethnic group is Turkish. Crucial role in the economy is agriculture, which employ 62.6% of the total number of employees in the municipality. In Kardzhali District largest number of cattle was in Kirkovo Municipality - 9865 and the number of sheep in the period 2001-2004 increased by 61.4% to 7710. From February till April 2014 volunteers from 4 villages – Samodiva, Shoptsi, Chorbadzhiisko and Benkovski **(Table 1)** participated in the screening.

Table 1. *General information of studied communities on CE in Kirkovo municipality, Kardzhali District*

Village	Population[1]	Ethnic[2]	Altitude[3]	Longitude	Latitude
Samodiva	284	Turkish	314	25.35°	41.367°
Shoptsi	391	Turkish	281	25.383°	41.383°
Chorbadzhiisko	1964	Turkish	331	25.4°	41.4°
Benkovski	2121	Turkish	380	25.267°	41.383°

[1]Population got from National Statistical Institute of Bulgaria according to the 2011 census
[2]Main ethnic groups in these villages
[3]Mean altitude sea level

Each person was registered (plus questionnaire) and screened by abdominal ultrasound. Two patients with liver CE cysts were detected by ultrasound. One of them was newly diagnosed 21 year old man with 7cm univesicular anechoic cystic lesion with double line sign in the right lobe. The other one was 79 year old woman with 10 cm relapsed multiseptated cystic lesion in the left lobe of the liver **(Table 2)**. They were subjected to further examination. The US prevalence of human CE was 0.3%. Of the questionnaire were found five previously diagnosed CE cases. There were 3 individuals with a history of surgery for CE that showed normal images at the time of the study **(Table 2).**

***Table 2**. Ultrasound findings, serology and other test results of the persons found to be CE positive*.

	Patients detected by US		Patient detected by questionnaire				
Years/ Gender	23/M	79/F	27/F	47/F	70/M	41/F	43/M
Village	Chorbadzhiisko	Shoptsi	Benkovski	Benkovski	Benkovski	Samodiva	Samodiva
Serology (ELISA)	Limit value	Positive	Limit value	Positive	Negative	Positive	Positive
X-ray	Non-specific	Non-specific	Non-specific	Non-specific	Non-specific	Non-specific	Non-specific
US	Univesicular anechoic cystic lesion with double line sign in the right lobe	Multiseptated cystic lesion in the left lobe of the liver	Normal	Normal	Normal	Cyst with detached membranes "water-lilly-sign"	Two cyst in the right lobe with heterogenous content. Univesicular anechoic cystic with double line sign in the spleen
Type of cyst	CE1	CE2	-	-	-	CE 3A	CE 4/ CE1
CT	Yes	No	No	No	No	No	Yes
Treatment	PAIR+ Albendazol	Patient refused treatment	Follow-up after surgery	Follow-up after surgery	Follow-up after surgery	Albendazol	Albendazol

In addition two persons have recently undergone treatment with albendazole for CE - one of them with liver localization, and the other with multiple cysts in the liver and spleen **(Table 2)**. Once of these cases we added additional data collected for the deceased after rupture of CE liver cyst 21 year old man from Shoptsi, the overall prevalence of CE in the present study was determined to be 1.25%. The youngest CE case was 23 yrs old and the average age of all 8 CE cases was 44 yrs (range 22-79). All of them are Turkish. In addition, three members of the family of the deceased man were examined and they were ultrasound and serologically negative for CE. All CE cases were analyzed for distribution and risk factors. The single factor logistic regression analysis showed that livestock

owner (OR=2. 73) and lack of dog deworming (OR=2. 38) was probably important risk factor for human CE in current study. The other relative risk factors might be age, dog owner, eating unwashed fruits and vegetables. Small sample size does not provide statistically significant relations **(Table 3)**. From all volunteers 50% (319/642) owned dogs of which 13.7 % owned dogs more than 5 yrs, and 10.2% - more than 10 yrs. 79 (25%) from dog owners said that they fed animal offal to dogs, and 141 (44.20%) have never deworming dogs. 551 (86%) from studied people have the habit to slaughter livestock at home. Only 196 (30.5%) people thought they knew something about echinococcosis. There were 218 (34%) who eat unwashed fruits and vegetables.

DISCUSSION

Although the National Programme for Control of Echinococcosis in humans and animals performed in the period 2004-2008 in Bulgaria (18), human CE cases still occurred (11). The total prevalence of confirmed human CE (new and existing cases) in Kirkovo municipality, Kardzhali District (East Rhodope Mountains, Bulgaria), was 1.25 (8/642) and ultrasound CE prevalence was 0.3% (2/642) in the current study. All CE cases were rural residents. The majority of population in the region are Muslim Turks who practice traditional home slaughtering of sheep and cattle. Those findings usually resulting from the presence of an active dog-sheep cycle, close contact with dogs and livestock, and human behavior, which contribute to main risk factors for human CE (19, 20). Local people's lifestyle around dogs and livestock might be the main reason for active *E. granulosus* transmission. Home slaughter was very common in Kardzhali district and a lot of persons reported to give row offals to their dogs. Poor knowledge of CE might have also contributed to the transmission likelihood over many years. From the above observation, CE

infection in humans and an active dog-sheep cycle appeared to be ongoing in Kirkovo municipality. Complex CE focused control program featuring physicians, veterinarians and local authorities should be implemented. In addition, the uncontrolled animal slaughter should be stopped, the dogs should not be fed with row organs and stray dogs should be kept under control, as well as is necessary to improve the knowledge of CE. Further portable ultrasound screening in the other municipalities of Kardzhali District must be continued.

***Table 3**. Univariable logistic regression analysis for risk factors of human CE in Kirkovo municipality.*

	Variables	Prevalence (%)	OR	95% Confidence Intervals CI
Gender	Male	4/ 302(1.32)	1.13	0.28 to 4.54
	Female	4/340(1,17)		
Ethnic	Turkish	8/629 (1.27)	0.40	0.02 to 7.20
	Others	0/14 (0.00)		
Occupation	Livestock owner	4/161 (2.48)	2.73	0.68 to 11.06
	Others	4/440 (0.9)		
Age (years)	< 19	0/ 128(0)		
	20-39	3/178(1,68)	1.60	0.38 to 6.76
	40-59	3/164(1,8)	1.79	0.42 to 7.55
	>60	2/182((1,09)	0.88	0.18 to 4.40
Dog owner	Yes	4/319 (1.25)	1.02	0.25 to 4.10
	No	4/324 (1.23)		
Lack of dog deworming	Yes	3/178 (1.75)	2.38	0.24 to 23.09
	No	1/141 (0.70)		
Eating unwashed	Yes	4/ 218(1,83)	1.96	0.49 to 7.91

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