

ECHINOCOCCOSIS

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HUMAN ECHINOCOCCOSIS

Human Hydatidosis / Hydatid disease

- Zoonotic parasitic infestation by a tapeworm of the genus *Echinococcus*



- Globally distributed
- WHO listed Echinococcosis among **17 neglected diseases** targeted for control or elimination by 2050 (Wen et al.,2019)

CLASSIFICATION

EMPIRE:- Eukaryota

PHYLUM:- Platyhelminthes

CLASS:- Cestoda

ORDER:- Cyclophyllidea

FAMILY:- Taenidea

GENUS:- *Echinococcus*



Species: 9

Species	Definitive host(s)	Intermediate host(s)	Human cases	Distribution
<i>Echinococcus granulosus sensu stricto</i>	Domestic dog, wolf, dingo, jackal, other canids	Sheep, goat, cattle, pig, camel, buffalo, horse, wild ungulates, marsupials, etc.	Yes	Cosmopolitan
<i>Echinococcus canadensis</i>	Domestic dog, wolf	Pig, camel, cervids	Yes	Eurasia, Africa, North and South America
<i>Echinococcus ortleppi</i>	Domestic dog	Cattle	Yes	Eurasia, Africa
<i>Echinococcus felidis</i>	Lion	Hyena, warthog, zebra, wildebeest, bush pig, buffalo, various antelopes, giraffe, hippopotamus	Not reported	Africa
<i>Echinococcus equinus</i>	Domestic dog	Horse, other equids, cervids	Not reported	Eurasia, Africa
<i>Echinococcus multilocularis</i>	All fox species, wolf, raccoon dog, domestic dog, cat	Arvicoline and microtine rodents and small herbivorous mammals, including lagomorphs (e.g., pika); pigs, boars, horses, cattle, nutrias, nonhuman primates, and dogs are accidental hosts	Yes	Eurasia, North America
<i>Echinococcus oligarthra</i>	Wild felids (e.g., <i>Puma concolor</i> [puma])	<i>Dasyprocta azarae</i> (agouti), <i>Didelphis marsupialis</i> (opossum)	Yes	Central and South America
<i>Echinococcus vogeli</i>	Bush dog, domestic dog	<i>Cuniculus paca</i> Linnaeus, 1766 (paca)	Yes	Central and South America
<i>Echinococcus shiquicus</i>	Tibetan fox	<i>Ochotona curzoniae</i> (Tibetan plateau pika)	Not reported	Tibetan Plateau

Fact

Exposure to food and water contaminated by the feces of an infected definitive host or poor hygiene in areas of infestation



Echinococcosis

Forms

- **Cystic Echinococcosis**, also known as Hydatid Disease or Hydatidosis or Unilocular Hydatidosis or dog tapeworm, caused by infection with a species complex centered on *E. granulosus*
- **Alveolar Echinococcosis**, also known as Multilocular Hydatidosis or alveolar hydatid disease or small fox tapeworm caused by infection with *E. multilocularis*
- **Neotropical Echinococcosis** (Two forms)
 - Polycystic caused by infection with *E. vogeli*; and
 - Unicystic caused by *E. oligarthrus*

History

- One of the oldest diseases known to man
- First described in the Talmud as a "Bladder full of water"
- Hippocrates described this disease more than two thousand years ago with a very interesting expression (**liver filled with water**)



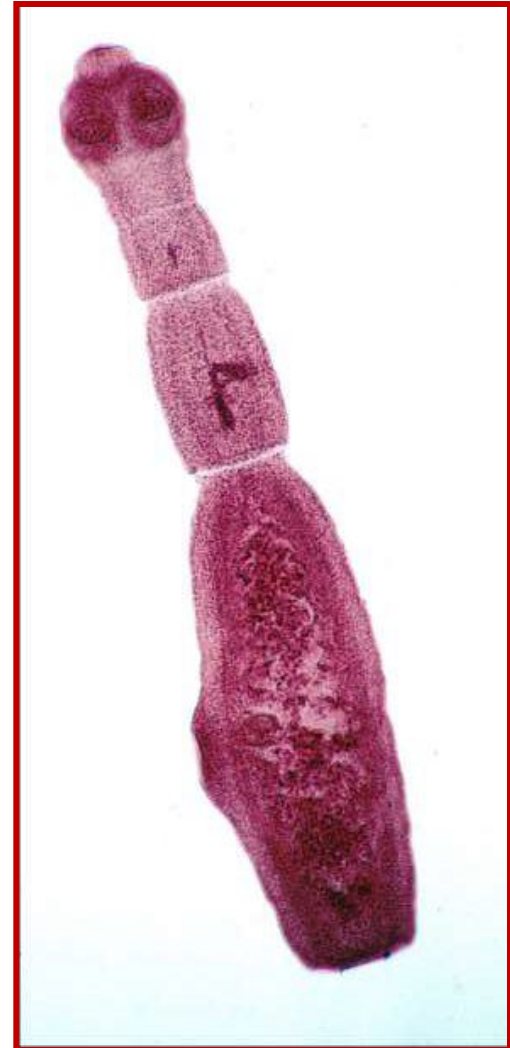
History

- ❖ Tyson (1683) – described hydatid in ‘rotten sheep’
- ❖ Hartmann (1695)- 1st described adult worm in intestine of dog
- ❖ Goeze (1782) – described scolices of cyst & indicated them as head
- ❖ Vogel (1957) *E. multilocularis* - confirmed it separate species
- ❖ Rausch & Bernstein (1972) *E. vogeli* - adult worm
- ❖ 1979 – larval stage 1st identified in surgical specimen from Columbia

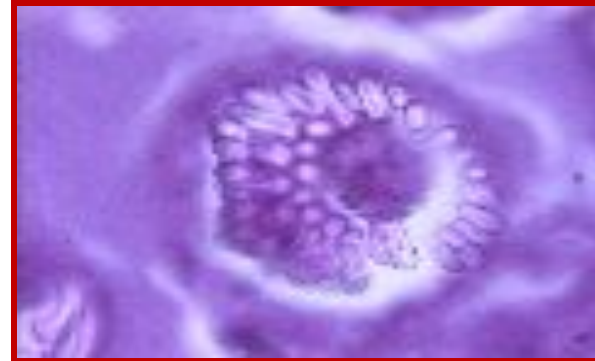
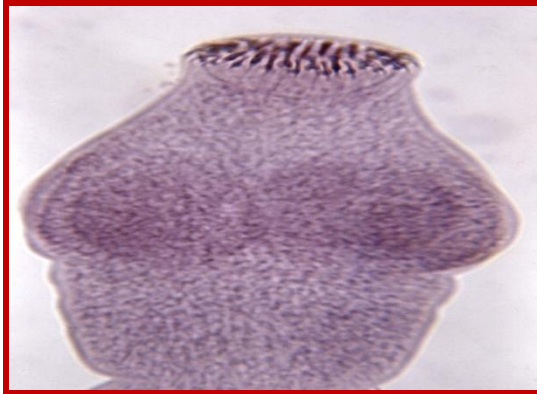
MORPHOLOGY

- **ADULT**
- **3-6 mm in length**
- 3-4 segments (proglottids)
- First Segment – Immature proglottid
- Second Segment- Mature proglottid
- Last segment (Biggest) – Gravid proglottid

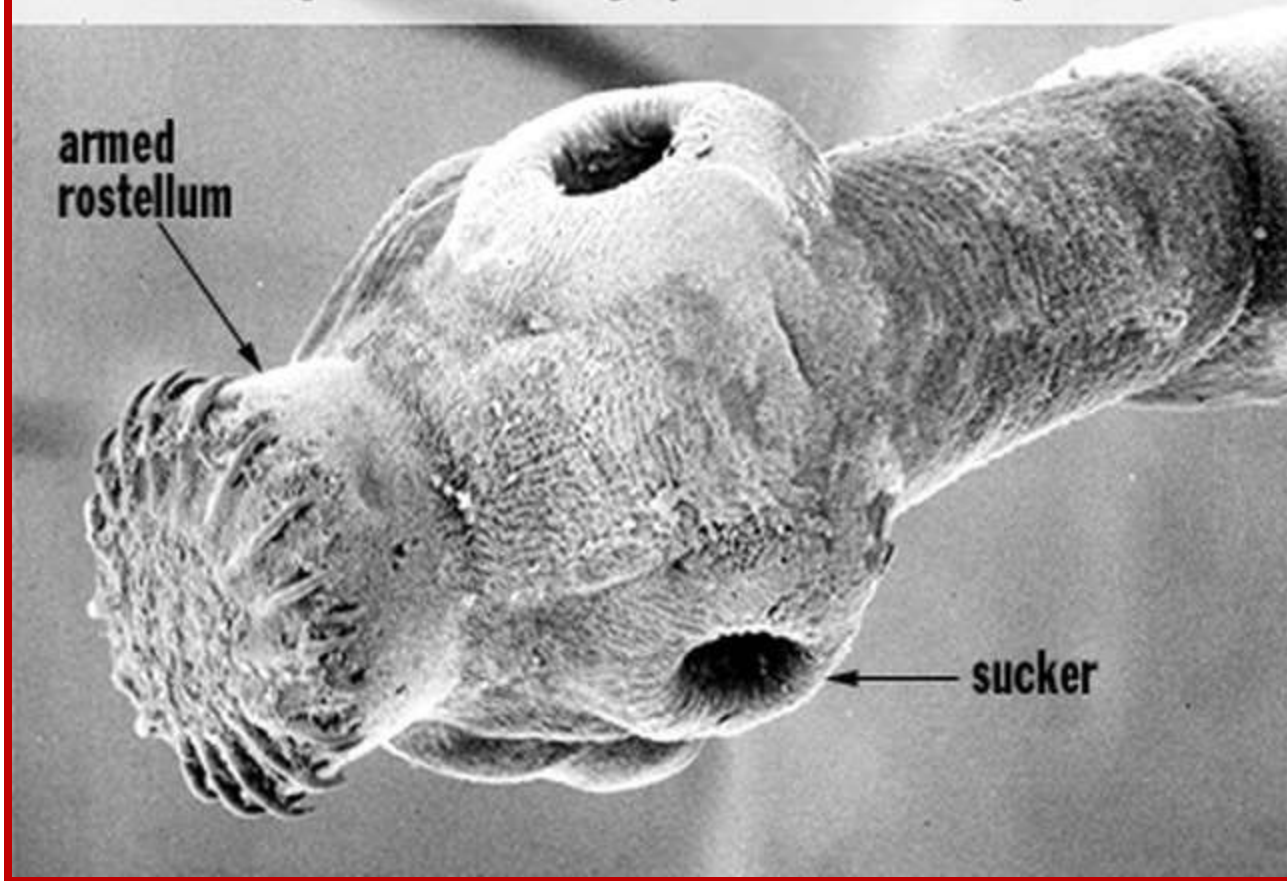
- Scolex (Head)
- Neck
- Strobila



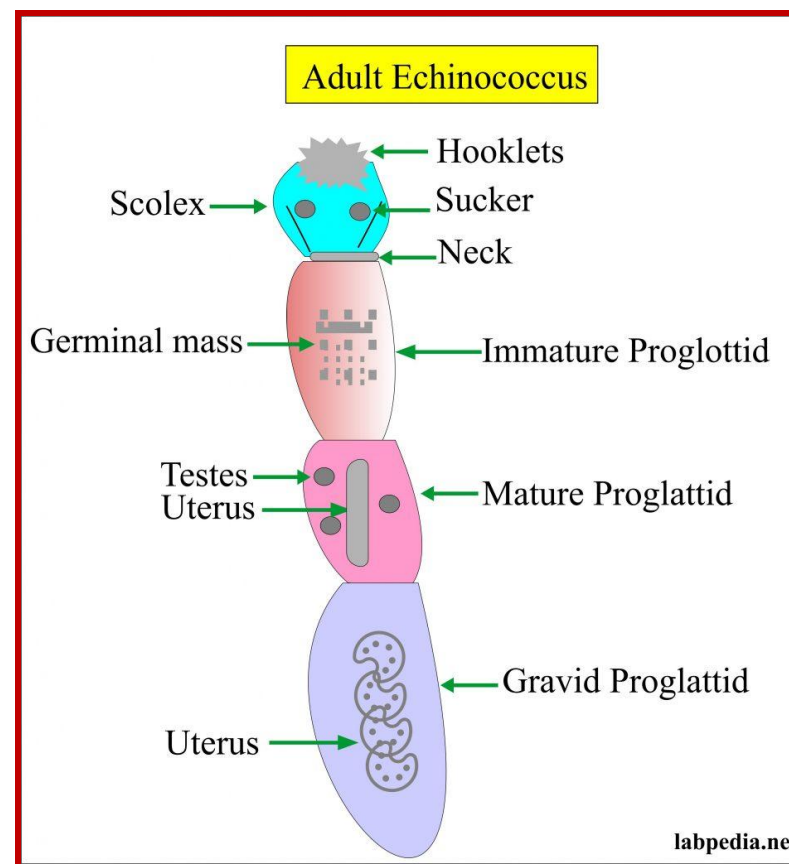
- **Scolex** –
 - 4 suckers
 - rostellum with double crown hooks.
 - Lack mouth/digestive cavity



Scanning electron micrograph: *Echinococcus* sp. adult



- **Neck**—
 - short, thick

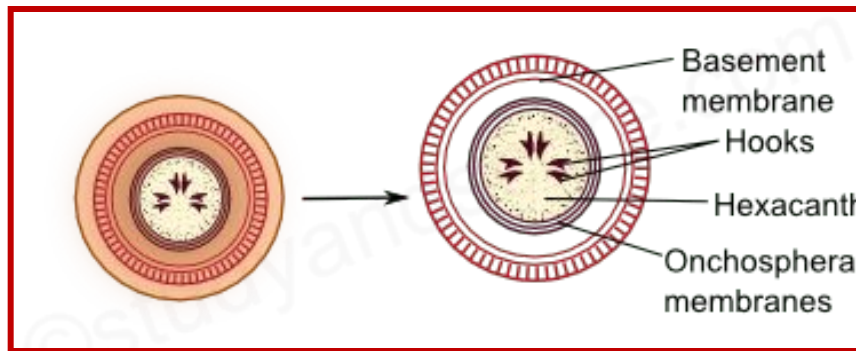


- **Strobila** - Proglottids
- **Posterior segment is gravid** with the uterus filled with eggs
- Average number of eggs / gravid proglottid is **823**



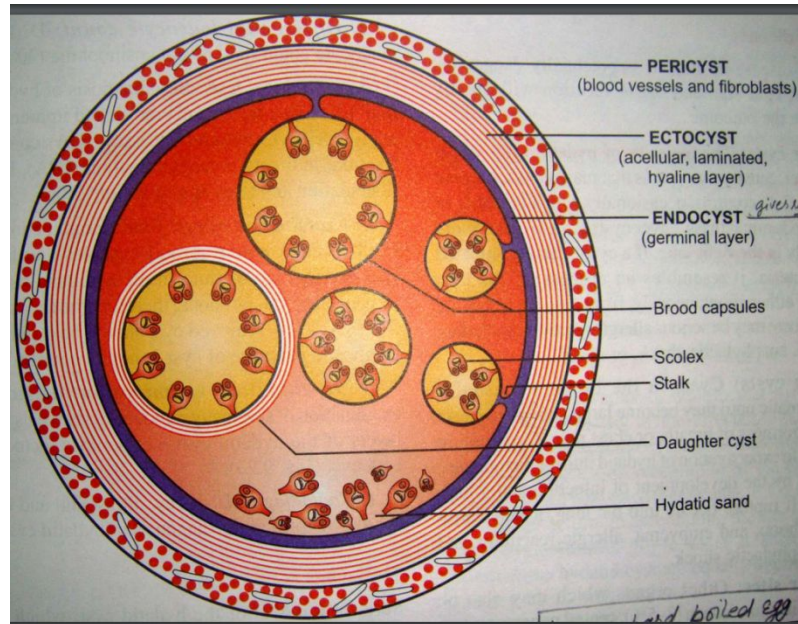
EGGS

- Ovoid
- Eggs contain an embryo that is called an oncosphere or hexacanth
- Hexacanth embryo with 3 pairs of hooks



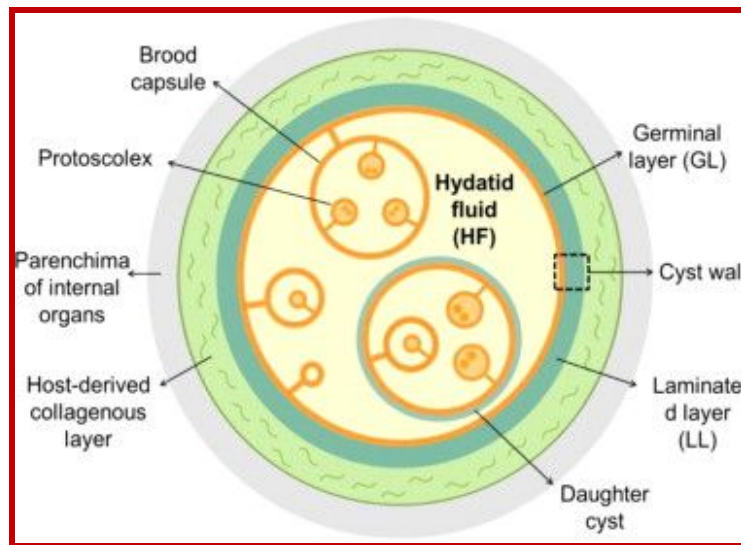
- **TWO LAYERS:**

- **ECTOCYST** : Acellular, 1mm thick (white of hard-boiled egg) Elastic so curls on itself when excised or ruptured



- **ENDOCYST** : Inner or Germinal layer 22-25 μm thick Gives rise to Ectocyst on outside and Brood capsules and Scolices on inside
- It secretes Hydatid Fluid Hydated Sand - When embryos breaks and float in fluid with in cyst

- Cystic structure filled with clear fluid (**hydatid fluid**)



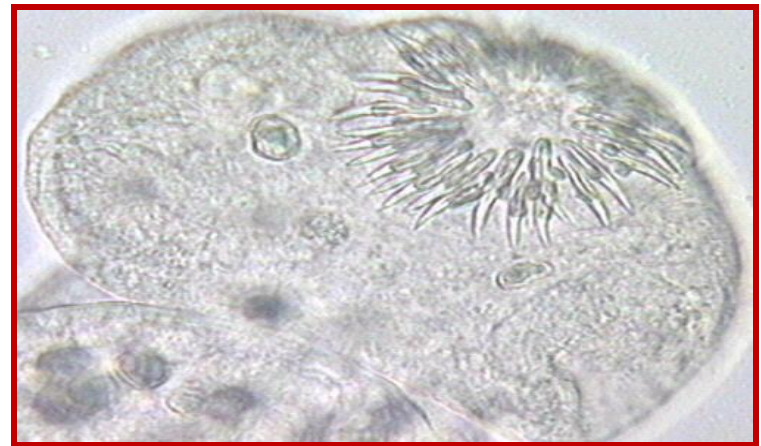
- Infective Stage
- Infective to man, cattle , sheep & other herbivorous animals

LARVAL FORMS

- Found within the hydatid cyst developing inside the intermediate hosts
- Represent the structure of the scolex of future adult worm
- On entering the definitive host, the scolex with four suckers and rostellar hooklets, becomes evaginated & develops into an adult worm

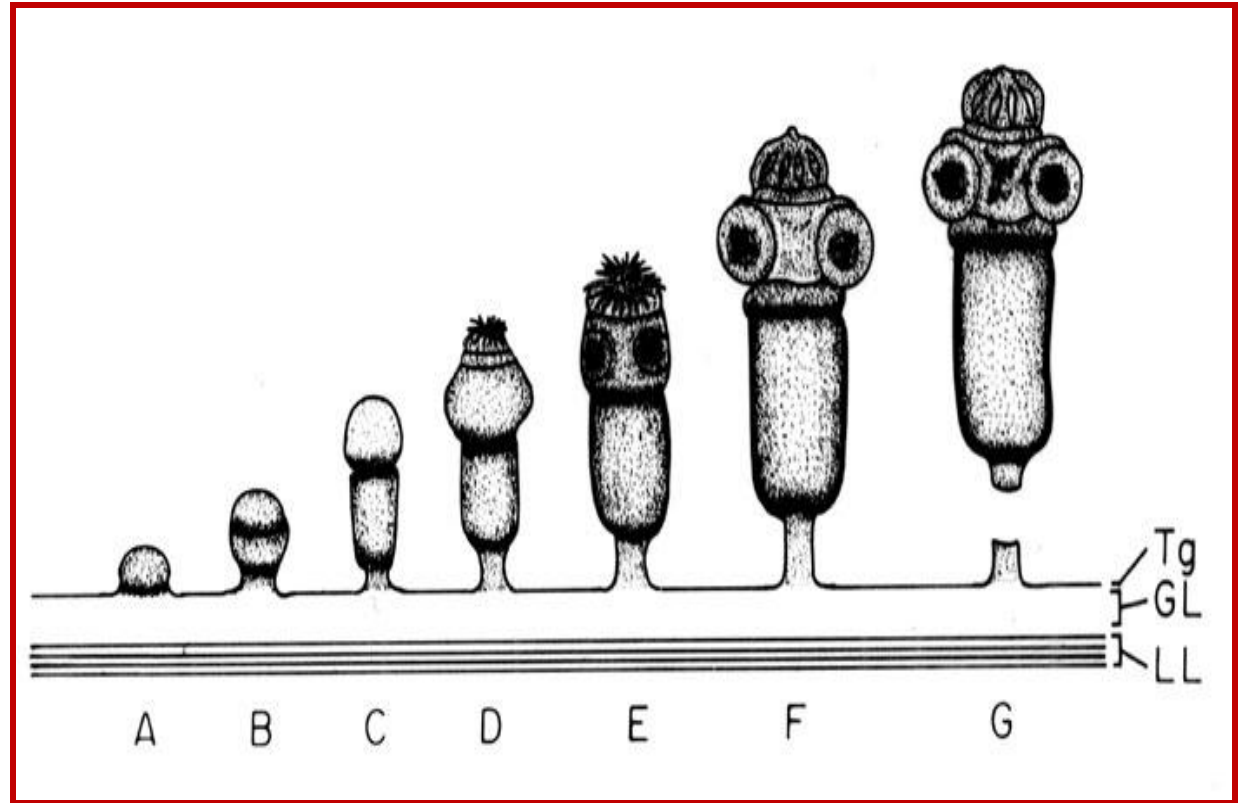


Developing Protoscolex



Mature Protoscolex

DEVELOPMENT IN HYDATID CYST



A-spherical bud

B-Early elongated bud

C-late elongated with presumptive scolex & body.

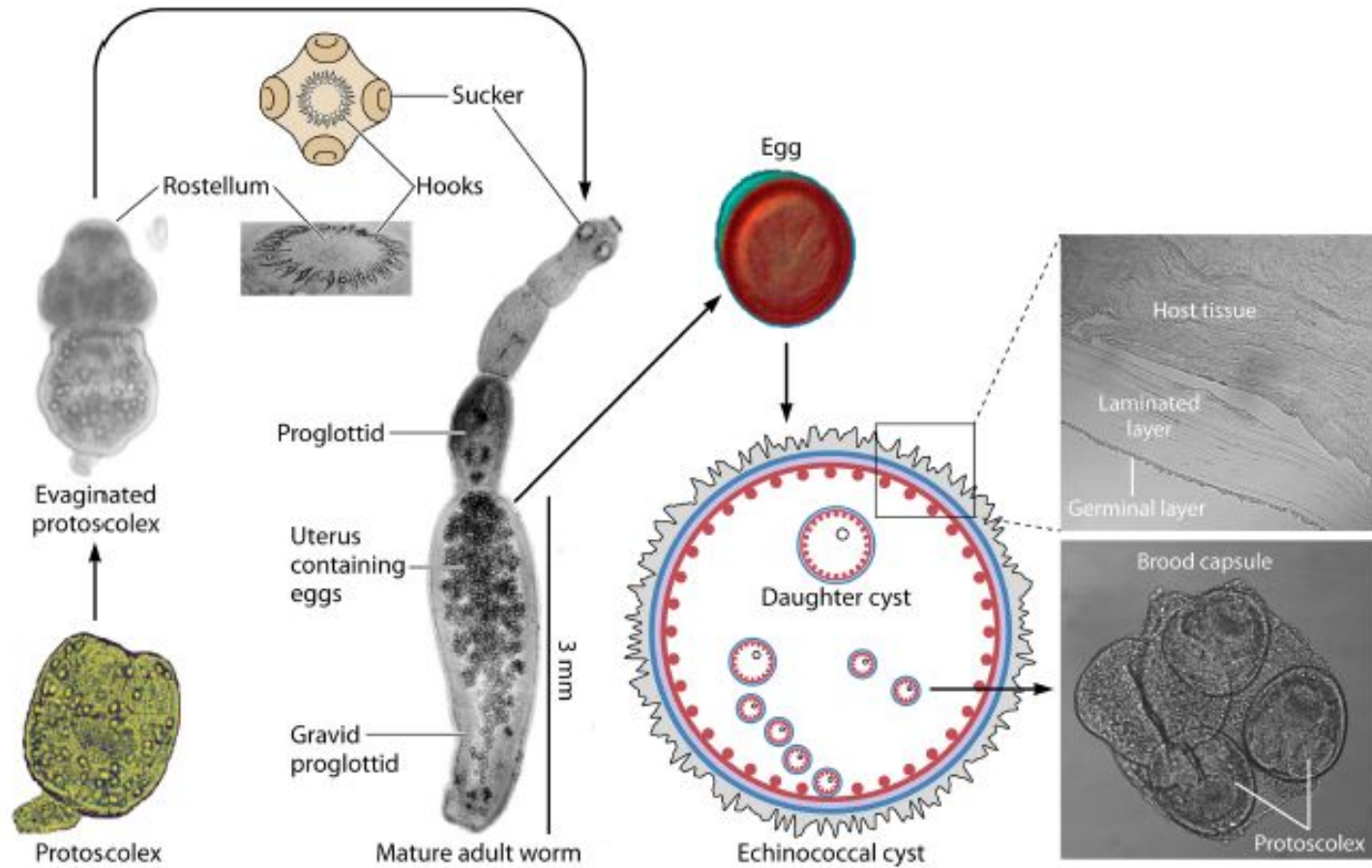
D- Protoscolex with rostellum formation

E-Protoscolex with suckers formation.

F-Fully developed protoscolex attached to germinal layer.

G-Protoscolex free in cyst cavity.

Echinococcus granulosus



Two most important forms, which are of medical and public health relevance in humans are

- ❖ **Cystic Echinococcosis (CE)**
- ❖ **Alveolar Echinococcosis (AE)**

Cystic Echinococcosis

caused by infection with the larval stage of *E. granulosus*

History



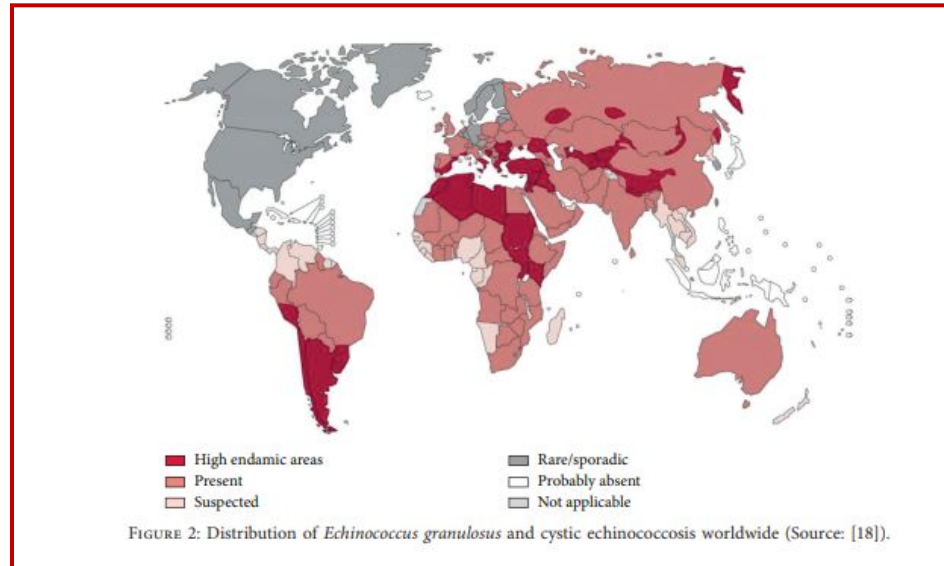
HIPPOCRATES

- *E. granulosus*- known since time of Hippocrates
- Hydatidosis (greek origin) - means **hedge hog berry**
- Hydatid : (greek origin) means **drop of water**



Distribution

- Globally distributed & found in every continent except Antarctic

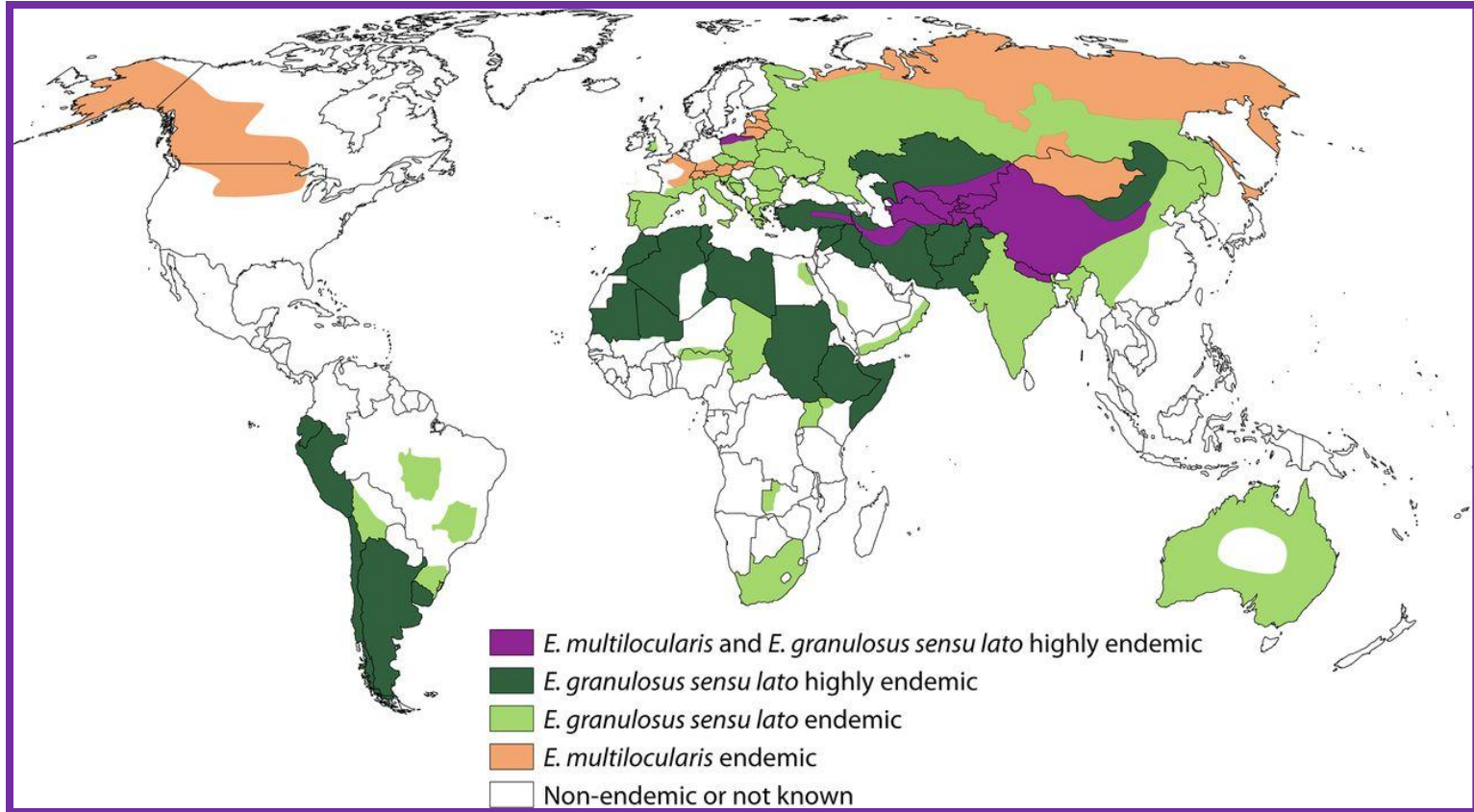


- In endemic regions, human incidence rates can reach more than 50 / 100 000 person-years
- Every year, disease is responsible for the loss of at least 1 million DALYs and of \$3 billion dollars in expenses, including treatment & livestock losses

- Women are affected more frequently than men; due to domestic activities that bring them in closer contact with dogs through feeding, herding, or milking livestock
- Prevalence of disease increases with age
- Prevalence levels as high as 5%–10% may occur in parts of Argentina, Peru, East Africa, Central Asia & China
- **India** has **the highest number** of CE cases (12% of global cases) worldwide (Kumar et al; 2020)
- This figure is likely to go up as the disease is often underreported in India

- In livestock, the prevalence found in slaughterhouses in hyperendemic areas varies from 20%–95% of slaughtered animals
- Highest prevalence is found - rural areas where older animals are slaughtered
- Involve reduction
 - carcass weight, decrease in hide value, decrease of milk production and reduced fertility

Epidemiology



Pattern of distribution has remained essentially unchanged over the past 2 decades

Epidemiology

- WHO: >1 million people affected globally each year
- Endemic area: annual CE incidence 1 to 200 per 100,000
- Mortality rate: 2% to 4%, may increase if inadequate management
- Eliminated from New Zealand
- Lack of accurate case recording prevents mapping of true epidemiological picture

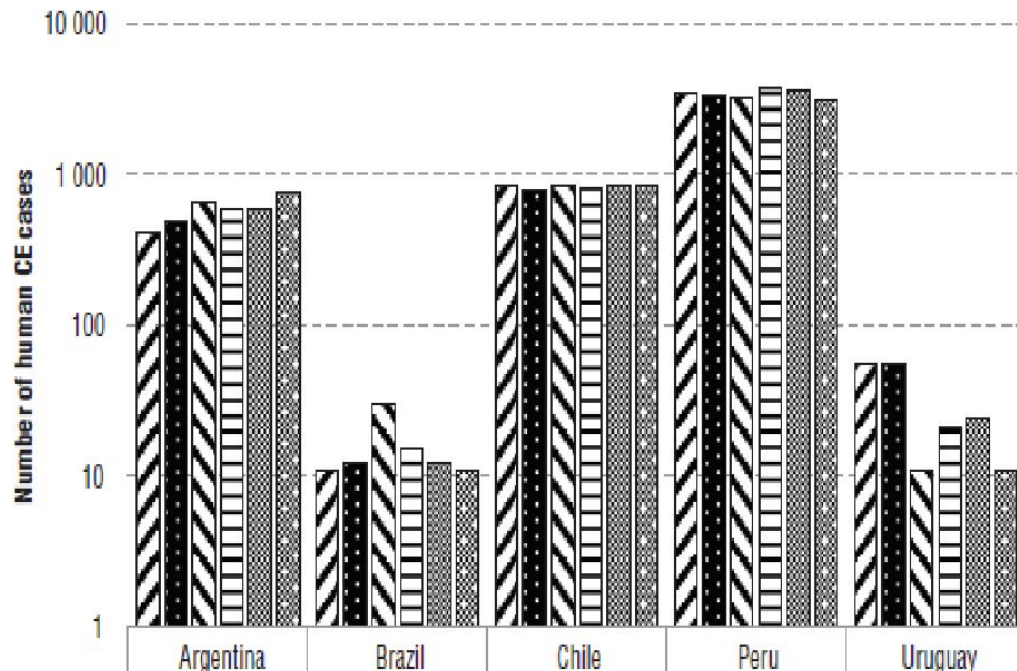
Cystic echinococcosis in South America: a call for action

Carlos F. Pavletic,¹ Edmundo Larrieu,² Eduardo A. Guamera,³ Natalia Casas,⁴ Pilar Irabedra,⁵ Ciro Ferreira,⁵ Julio Sayes,⁵ Cesar M. Gavidia,⁶ Eduardo Caldas,⁷ Michael Laurence Zini Lise,⁷ Melody Maxwell,⁸ Marcos Arezo,⁹ Ana Maria Navarro,¹⁰ Marco A. N. Vigilato,¹¹ Ottorino Cosivi,¹¹ Marcos Espinal,¹¹ and Victor J. Del Rio Vilas¹¹

Rev Panam Salud Publica 41, 2017

- Nearly 5000 new CE cases were diagnosed annually in the five countries during the study period

FIGURE 1. Number of cases of cystic echinococcosis (CE) in humans in South America, by country, 2009 – 2014



	Argentina	Brazil	Chile	Peru	Uruguay
2009	416	11	843	3 556	57
2010	496	12	799	3 413	55
2011	670	30	832	3 275	11
2012	598	15	815	3 784	21
2013	585	12	838	3 625	24
2014	777	11	832	3 132	11

Epidemiology of echinococcosis in Iran: a systematic review and meta-analysis

[Shima Mahmoudi](#), [Setareh Mamishi](#), [Maryam Banar](#), [Babak Pourakbari](#) & [Hossein Keshavarz](#) 

[BMC Infectious Diseases](#) **19**, Article number: 929 (2019) | [Cite this article](#)

- From 1 January 1990 to 8 August 2017
- Pooled prevalence: 5%
- Patients younger than 40 years age

Seroprevalence of human cystic echinococcosis from North India (2004-2015)

[Kamran Zaman](#)¹, [Abhishek Mewara](#)¹, [Sunil Kumar](#)¹, [Kapil Goyal](#)¹, [Sumeeta Khurana](#)¹, [Praveen Tripathi](#)¹, [Rakesh Sehgal](#)¹

¹Department of Medical Parasitology, Postgraduate Institute of Medical Education and Research, Chandigarh, India.

Materials and methods: A retrospective analysis of laboratory data of 3929 clinically and/or radiologically suspected cases of CE was carried out for 12 years from 2004 to 2015 and compared to the previous data from 1984 to 2003. The seroprevalence of anti-hydatid immunoglobulin G (IgG) was assessed by enzyme-linked immune sorbent assay. Casoni's intradermal skin test and microscopy on aspirated hydatid cyst fluid were also done. The statistical significance was assessed using Chi-square test and Fisher's *t*-test.

Results: Of the 3929 samples, 1124 (28.6%) were positive for specific anti-hydatid IgG antibody response, while of the 121 tested by Casoni's test, 56 (46.3%) were positive. The seropositivity of CE over the period of 12 years is rising. As compared to our previous data from 1984 to 2003, an overall significant increase in seropositivity was observed during 2004-2015 (28.6% vs. 15.0% in 1984-2003, $P < 0.0001$).

Conclusions: This study emphasizes the necessity of continuous surveillance and integrated control measures to prevent CE in humans and livestock across the country.

Sero-Epidemiological Survey of Human Cystic Echinococcosis in Kashmir, North India

Bashir Ahmad Fomda^{1*}, Asiya Khan¹, Manzoor Ahmad Thokar¹, Ajaz Ahmad Malik², Anjum Fazili³, Rayees Ahmad Dar⁴, Monika Sharma⁵, Nancy Malla⁵

1 Department of Microbiology, Sher-i-Kashmir Institute of Medical Sciences, Srinagar, Jammu and Kashmir, India, 2 Department of General Surgery, Sher-i-Kashmir Institute of Medical Sciences, Srinagar, Jammu and Kashmir, India, 3 Department of Community Medicine, Sher-i-Kashmir Institute of Medical Sciences, Srinagar, Jammu and Kashmir, India, 4 Department of Biostatistics, Sher-i-Kashmir Institute of Medical Sciences, Srinagar, Jammu and Kashmir, India, 5 Department of Medical Parasitology, Post Graduate Institute of Medical Education and Research, Chandigarh, India

Risk Factors	N (Seropositive/total) (%)	RR (95% of CI)	P-value*
Gender			
<i>Male</i>	42/630 (6.6)	1.831(1.132–2.961)	0.012
<i>Female</i>	30/799 (3.75)		
Residence			
<i>Rural</i>	70/1161(5.26)	8.533(2.079–35.020)	<0.001
<i>Urban</i>	02/268 (4.59)		
Age group			
<i>1–15 years</i>	19/148 (5.79)	Reference	
<i>16–55 years</i>	41/1015 (4.07)	3.643 (2.052–6.467)	<0.001
<i>Above 55 Years</i>	12/266 (3.05)	3.118 (1.468–6.621)	0.002
Contact with Dog			
<i>Contact</i>	62/965 (6.42)	3.11 (1.583–6.137)	<0.001
<i>No contact</i>	10/464 (2.15)		

- Out of 1,429 samples, 72 (5.03%) IgG positive by ELISA

Review Article

Human Cystic Echinococcosis with Special Reference to India – An Overview

Nancy Malla* and Abhishek Mewara

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Geographical area(s)	Positive numbers
Chandigarh, North Indian states (Gochhaitt et al)	8 (6.4%) in 2011-2013
Central and southern Andhra Pradesh, South India (Khader et al)	118 CE patients in 2009- 2011
West Bengal, East India (Ghoshal et al)	106 in 2005-2009
Maharashtra, Central India (Akther et al)	117 in 1996-2007
Andhra Pradesh, South India (Hemachander et al)	11 (11%) in November 2005 – May 2006
Chandigarh, North Indian states (Khurana et al)	495 (15%) seropositive in 1984 to 2003
Chandigarh, North Indian states (Bakshi et al)	1 (5.8%) during 1999-2004
Kashmir Valley, North India (Fomda et al)	705 (12.1%) during 1984-2001



Chapter Six - Global Distribution of Alveolar and Cystic Echinococcosis

P. Deplazes ^{*}, L. Rinaldi [§], C.A. Alvarez Rojas [¶], P.R. Torgerson ^{*}, M.F. Harandi ^{||}, T. Romig [#], D. Antolova ^{**}, J.M. Schurer ^{§§}, S. Lahmar ^{|||}, G. Cringoli [§], J. Magambo ^{###}, R.C.A. Thompson ^{***}, E.J. Jenkins ^{§§}

Country	Human	Canids	Cervids	Swine	Country	Human	Dog	Sheep	Cattle	Goat (Go)/Alpaca (A)	Pig
Canada		G8 ^{1,2} G10 ^{1,2,3,4}	G8 ^{4,5} G10 ^{4,5}		Argentina	G1 ^{1,2,3,4} G2 ^{2,4} G5 ² G6 ^{2,4}	G1 ^{3,5} G6 ⁵	G1 ^{4,5} G2 ⁴ G3 ⁵	G1 ^{1,3}	Go: G1 ⁵ Go: G6 ⁵	G1 ³ G7 ^{4,5}
United States	G8 ⁶		G8 ^{4,7} G10 ⁴	Historically present but eradicated	Brazil	G1 ⁶ G3 ⁶ G5 ⁶	G1 ⁶ G3 ⁶ G5 ⁶	G1 ⁸	G1 ^{3,7,8} G5 ^{3,7,8}		G1 ⁹ G7 ⁹
México	G5 ⁸	G7 ⁹		G1 ¹⁰ G7 ^{9,10,11}	Chile	G1 ^{3,10} G6 ¹⁰			G1 ^{3,11} G3 ¹¹		
					Peru	G1 ^{12,13,14} G6 ^{12,14}		G1 ^{12,13}	G1 ^{12,13}	A: G1 ¹⁵ Go: G6 ¹²	G1 ¹⁵ G7 ^{12,15}
					Uruguay				G1 ^{1,3} G5 ¹		

Genotypes of *Echinococcus* spp. causing cystic echinococcosis in North America

Table 5 Genotypes of *Echinococcus* spp. causing cystic echinococcosis in South America: *Echinococcus granulosus* (G1–3), *Echinococcus ortleppi* (G5), *Echinococcus intermedius* (G6/7)

Table 6 Genotypes of *Echinococcus* spp. causing cystic echinococcosis in eastern Central Europe: *Echinococcus granulosus* (G1–3), *Echinococcus equinus* (G4), *Echinococcus ortleppi* (G5), *Echinococcus intermedius* (G6/7) and *Echinococcus canadensis* (G8, G10) (no data found in the missing countries)

Country	Human	Dog	Wild canids	Cattle	Pig (P), Wild boar (Wb)	Sheep (S), Cervids (C)
Poland	G7 and G1 ¹				P: G7 ³	
Estonia		G1 ²	G8 ² G10 ² G10 ²			C: G8 and G10 ^{2,4}
Latvia						
Lithuania	G7 ⁵	G7 ^{2,5}		G7 ^{2,5}	P: G7 ^{2,5}	
Ukraine					P, Wb: G7 ⁶	
Moldova				G1 ⁷ G3 ⁷		S: G1 ⁷ S: G3 ⁷
Slovakia	G7 ⁸ and G1–3*				P: G7 ⁸	

Prevalence of *E. granulosus* in INDIA

- As per 20th livestock census, 2019, total dog population - 9.43 million in India
- According to other animal welfare groups & websites, country is home to about 30 million stray dogs, which amounts to 1 stray dog / 42 people (Socialcops, 2020; Statista, 2020)



Epidemiology, Risk Factors and Economics of Echinococcosis in India: A Review

Kushal Grakh, Anand Prakash, Dinesh Mittal*, Pankaj Kumar and Ramesh Kumar

Department of Veterinary Public Health and Epidemiology, Lala Lajpat Rai University of Veterinary and Animal Sciences, Hisar, Haryana, INDIA

Table: Prevalence (%) of *E. granulosus* in stray dog population in India

Location/State	Prevalence	Reference
Kurnool, Andhra Pradesh	33.3	Reddy <i>et al.</i> , 1958
Delhi and Uttar Pradesh	14	Singh and Dhar, 1988
Assam	17.02	Deka <i>et al.</i> , 2008
Mizoram	18.18	Deka <i>et al.</i> , 2008
Meghalaya	27.77	Deka <i>et al.</i> , 2008
Bangalore urban, Karnataka	4.35	Prathiush <i>et al.</i> , 2008
Maharashtra	5.22 - 6.57	Nikale <i>et al.</i> , 2014; Ingole <i>et al.</i> , 2018
Gurugram, Haryana	13.5	Varma, 1990

Region	Prevalence					Literature
	Cattle	Buffalo	Sheep	Goat	Pig	
Northern India						
Haryana			4.7	1.9	1.3	Varma, 1990
Punjab	7.6	8.9-50.96	5.1	2.8	4.66	Deka <i>et al.</i> , 1983, Sharma <i>et al</i> 2004, Jadhav <i>et al.</i> , 2013, Khan <i>et al.</i> , 2013
HP			7.2	2.3		Jitendran, 1996
J&K				19.8		Godara <i>et al.</i> , 2014
Uttar Pradesh		14.82- 36	2.56-6	1.45-2	0.9-1.42	Deka and Gaur 1998, Varma and Malviya 1992, Irshadullah <i>et al.</i> , 1989; Ganaie <i>et al.</i> , 2018
Delhi			18.5			Rana <i>et al.</i> , 1986
Diff. Location	5.39	4.36	2.23	0.41	3.09	Singh <i>et al.</i> , 2014b
Diff. Location		48.1	30.50%	21		Singh and Dhar, 1988
Range	5.4-7.6	4-51	2.2-30	0.41-21	0.9-4.6	
Western India						
Maharastra	5.1	3.81	0.02-1.06	0.3	0.87	Pednekar <i>et al.</i> , 2009, Gatne 2001
Rajasthan			6.57	4.12		Shekhawat <i>et al.</i> , 2005
MP		1.19				Jatav and Garg, 2012
Range	5.1	1.2-3.8	0.02-6.6	0.3-4.1	0.9	
Southern India						
AP			5.18-7.05	1.39		Pillai <i>et al.</i> , 1986; Hafeez <i>et al.</i> , 1994
Chennai			5.6-22.4	7-25.8		Sangaran <i>et al.</i> , 2014; Shanmugam <i>et al.</i> , 1994
Karnataka			0.37-4.9	4.78		Vijayasmitha <i>et al.</i> , 1993
Kerala	35.47	33	2.55	1.05		Abraham <i>et al.</i> , 1980
Diff. Location	7.1	9.4	7		11.5	Hafeez,1997
Range	7.1-35.4	9.4-33	0.4-22.4	1.0-25.8	11.5	
Eastern India						
Assam	16	6.52		2.24-4.9	0.43-1.8	Sarma <i>et al.</i> , 2000; Deka <i>et al.</i> , 2008
Bihar				8.48	7.6-8.25	Kumar <i>et al.</i> , 2007; Prasad, 1981
Meghalaya	21.40%				0.34	Deka <i>et al.</i> , 2008
Pudduchery			47.6	37.8		Das and Sreekrishnan, 1998
Sikkim			50	33.3		Katiyar and Sinha, 1981
West Bengal			10.5	3.9	8	Biswas <i>et al.</i> , 1989; Das and Das, 1998
Range	16-21.4	6.5	10.5-50.0	2.2-57.8	0.3-8.3	
Overall	1.6-35.4	1.2-51	0.02-50	1.05-57.8	0.3-11.5	

Table : Zone and state wise prevalence (%) of CE in intermediate hosts

Life Cycle

The worm passes its life cycle in two hosts

(1) **Adult tapeworm** in the **Definitive host**

- Dogs (Optimum Definitive Host)
- Wolf
- Fox
- Jackal



The adult worm lives in the small intestine in these animals who discharge a large number of eggs

(2) **Eggs** in the environment which are swallowed by the **Intermediate hosts**

- Sheep (Optimum Intermediate Host)
- Pig
- Cattle
- Horse
- Goat
- Man (act as an accidental intermediate host)

The larval stage is passed in these animals and man giving rise to hydatid cyst

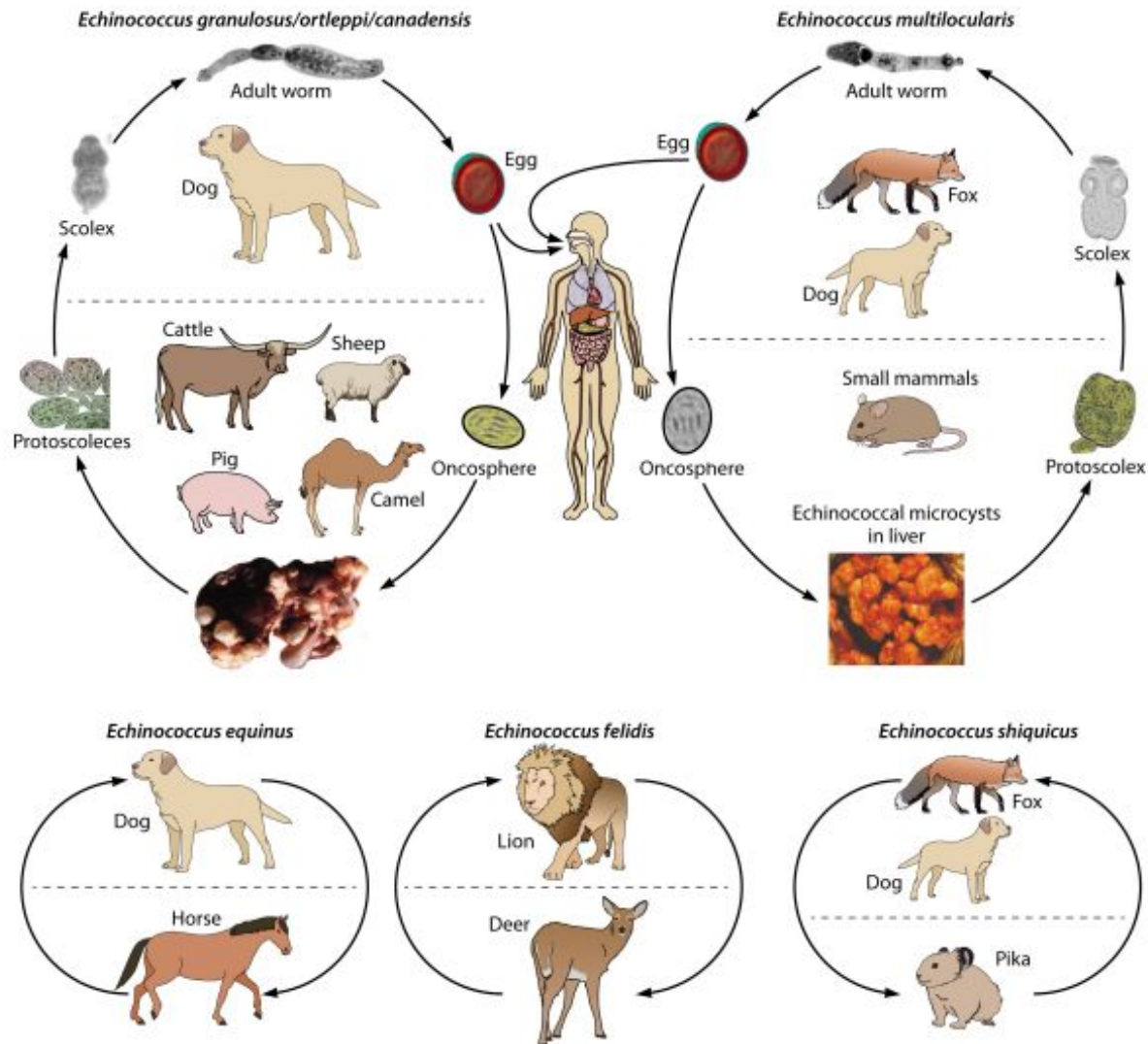


FIG 1 Life cycles of *Echinococcus* spp. Species responsible for human infection (*E. granulosus sensu stricto*, *E. ortleppi*, and *E. canadensis* [belonging to *E. granulosus sensu lato*] and *E. multilocularis*) are shown at the top. Species at the bottom (*E. shiquicus*, a species close to *E. multilocularis*, and *E. equinus* and *E. felidis*, belonging to *E. granulosus sensu lato*) are not known to cause disease in humans. Only the most common definitive and intermediate hosts which play a major role in life cycle/transmission are shown; other hosts may be encountered (especially wildlife hosts for *E. granulosus sensu lato* and domestic hosts for *E. multilocularis*). *E. vogeli* and *E. oligarthra*, which are responsible for polycystic echinococcosis in humans in Central and South America, are not represented in the figure.

Risk Factors for the Disease

- Suitable conditions for the establishment & transmission exist in India
 - Large population of intermediate hosts
 - Close contact of human-dog and dog and intermediate host
 - Browsing and grazing on contaminated pasture
 - Warm & humid and cold climate conditions during different part of seasons (Singh et al., 2014)

- Livestock population of 536.76 million, out of which 536.16 million can act as intermediate hosts to complete the life cycle (20th Livestock Census, 2019)
- Intermediate hosts graze on pastures contaminated with dog faeces & ingest echinococcal eggs
- High number of stray dog population in both urban & rural areas in India is another risk factor responsible for higher prevalence of *E. granulosus*

- These **stray dogs** receive no or an inadequate treatment with anti-parasitic medications - play an important role in the transmission of the disease
- Open dumping of dead animals in rural parts - with an easy access to stray dogs further complicate the problem
- Pet dogs as guarding animals, dog owners, animal handlers, veterinarians & laboratory workers are also at higher risk of infection, since the eggs can contaminate water, fruits & vegetables

Molecular epidemiology of *Echinococcus species*

- Mitochondrial DNA sequences have identified **10** different genetic types of *E. granulosus* from G1 to G10 (Craig et al., 2007)
- Livestock from **Eastern India** demonstrated the presence of four genotypes
 - G1 (sheep strain)
 - G2 (Tasmanian sheep strain)
 - G3 (buffalo strain)
 - G5 (cattle strain) (Bhattacharya et al., 2007)

- In **Northern India**, 77.7% of isolates from cattle, pig, buffalo and goat were grouped under G3 strain and 22.2% isolates from sheep are grouped under G1 strain (Singh et al., 2012)
- Four different genotypes i.e. G1, G2, G3 and G5 are demonstrated in food producing animals viz. cattle, buffalo, sheep and pigs in **Western India** (Pednekar et al.,2009)
- Indicate that **G3** is the most common strain of *E. granulosus* circulating in livestock in India followed by G5, G1 and G2 strain

Molecular epidemiology of *Echinococcus species*

- Despite the isolation of G5 and G6 in humans, G1 and G3 remained commonly encountered strains in human CE in India (Sharma et al., 2013)
- Strains of *Echinococcus* spp. circulating in livestock in India are well adapted to cause infection in all the intermediate hosts of *E. granulosus*

- **Globally**, most of the human cases of CE are found to be infected with G1 strain of *E. granulosus* (Moro and Schantz, 2009)
- **G1** genotype of *Echinococcus granulosus* is responsible for the vast majority (88%) of human cases worldwide
- It has a cosmopolitan distribution and is associated with transmission from sheep as an intermediate host
- Wide variation in *E. granulosus* strains may influence the life-cycle patterns, development rate, specificity to host, transmission, antigenic proteins in fluid and response to treatment (Craig et al., 2007).

Genetic Diversity and Population Genetic Structure Analysis of *Echinococcus granulosus sensu stricto* Complex Based on Mitochondrial DNA Signature

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- Hydatid cysts collected randomly during 2010-2012 from 4 different geographical areas in North India
- Total isolates from 81 animals analyzed
- 3 genotypes found: G3 genotype (n=58), G1 genotype (n=22) & G2 genotype (n=1)

Molecular Characterization of *Echinococcus granulosus* Cysts in North Indian Patients: Identification of G1, G3, G5 and G6 Genotypes

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Abstract

Background: Cystic echinococcosis (CE) caused by the *Echinococcus granulosus*, is a major public health problem worldwide, including India. The different genotypes of *E. granulosus* responsible for human hydatidosis have been reported from endemic areas throughout the world. However, the genetic characterization of *E. granulosus* infecting the human population in India is lacking. The aim of study was to ascertain the genotype(s) of the parasite responsible for human hydatidosis in North India.

Methodology/Principal Findings: To study the transmission patterns of *E. granulosus*, genotypic analysis was performed on hydatid cysts obtained from 32 cystic echinococcosis (CE) patients residing in 7 different states of North India. Mitochondrial cytochrome *c oxidase* subunit1 (*cox1*) sequencing was done for molecular identification of the isolates. Most of the CE patients (30/32) were found to be infected with hydatid cyst of either G3 (53.1%) or G1 (40.62%) genotype and one each of G5 (cattle strain) and G6 (camel strain) genotype.

Conclusions/Significance: These findings demonstrate the zoonotic potential of G1 (sheep strain) and G3 (buffalo strain) genotypes of *E. granulosus* as these emerged as predominant genotypes infecting the humans in India. In addition to this, the present study reports the first human CE case infected with G5 genotype (cattle strain) in an Asian country and presence of G6 genotype (camel strain) in India. The results may have important implications in the planning of control strategies for human hydatidosis.

Citation: Sharma M, Sehgal R, Fomda BA, Malhotra A, Malla N (2013) Molecular Characterization of *Echinococcus granulosus* Cysts in North Indian Patients: Identification of G1, G3, G5 and G6 Genotypes. PLoS Negl Trop Dis 7(6): e2262. doi:10.1371/journal.pntd.0002262

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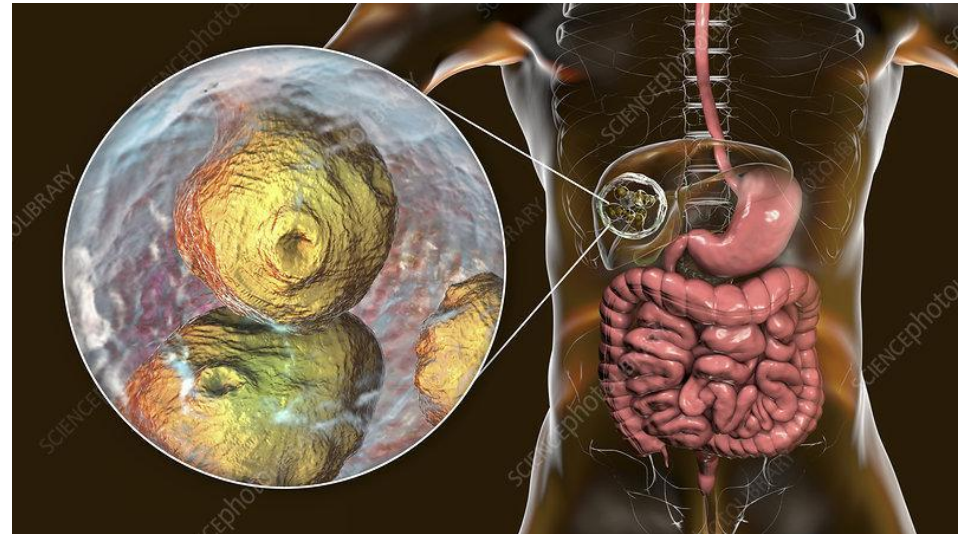
* E-mail: drmallanancy@gmail.com

CLINICAL FEATURES

- Leads to the development of one or more hydatid cysts located most often in the liver and lungs, and less frequently in the bones, kidneys, spleen, muscles and central nervous system

Organs affected by *E granulosus* are:

- Liver (63%)
- Lungs (25%)
- Muscles (5%)
- Bones (3%)
- Kidneys (2%)
- Brain (1%)
- Spleen (1%)



- Asymptomatic incubation period of the disease can last many years until hydatid cysts grow to an extent that triggers clinical signs

Spine Echinococcosis

- The incidence of osseous echinococcosis is low (approximately 0.5–4%)
- Spinal involvement is the most common form, though rare overall (0.2– 1%)
- The most common spinal location is the thoracic spine (approx 50%), followed by the lumbosacral region (approx 29%) and the lumbar spine (approx 21%)
- Hydatid cysts are usually not confined to the vertebral bodies, often, they affect the intervertebral discs, the spinal cord and the posterior spinal elements, and they may grow in the spinal canal as well

CLINICAL FEATURES

- If the **liver** is affected
 - Abdominal distention & pain, nausea and vomiting, upper abdominal discomfort and poor appetite; compression of bile ducts may lead to jaundice
- If the **lung** is affected :
 - chronic cough, chest pain and shortness of breath
- Other signs depend on the location of the hydatid cysts and the pressure exerted on the surrounding tissues
- Non-specific signs include
 - anorexia, weight loss and weakness

CLINICAL FEATURES

- If there is **Cerebral involvement**
 - Headache
 - Dizziness
 - Decreased level of consciousness
- **In bone infections**
 - Occur and produce thin and fragile bones, that can lead to spontaneous fracture

Spinal echinococcosis

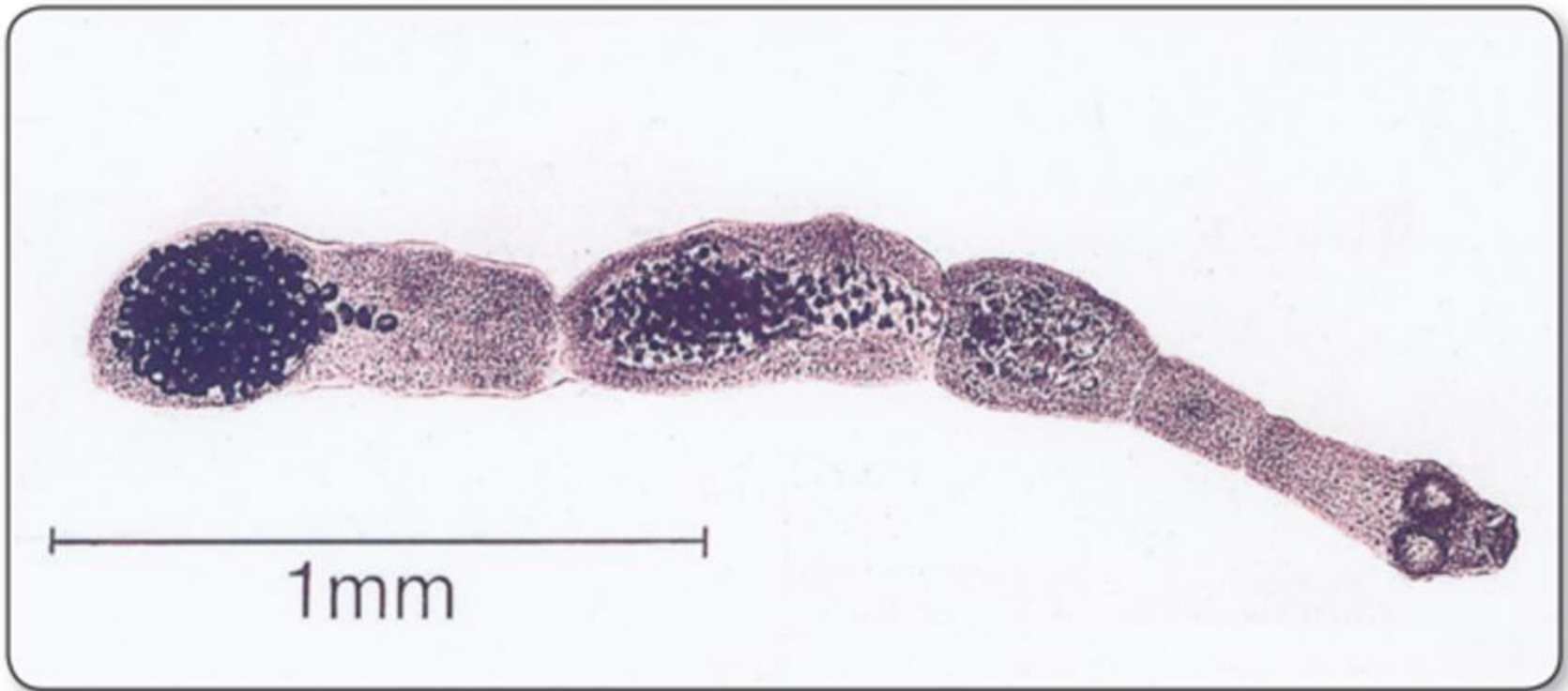
stemming from cyst compression of adjacent spinal structures. The patients most commonly present with back pain; limb weakness occurs later.

Other presenting symptoms include radiculopathy, myelopathy and pathological fractures

Alveolar Echinococcosis

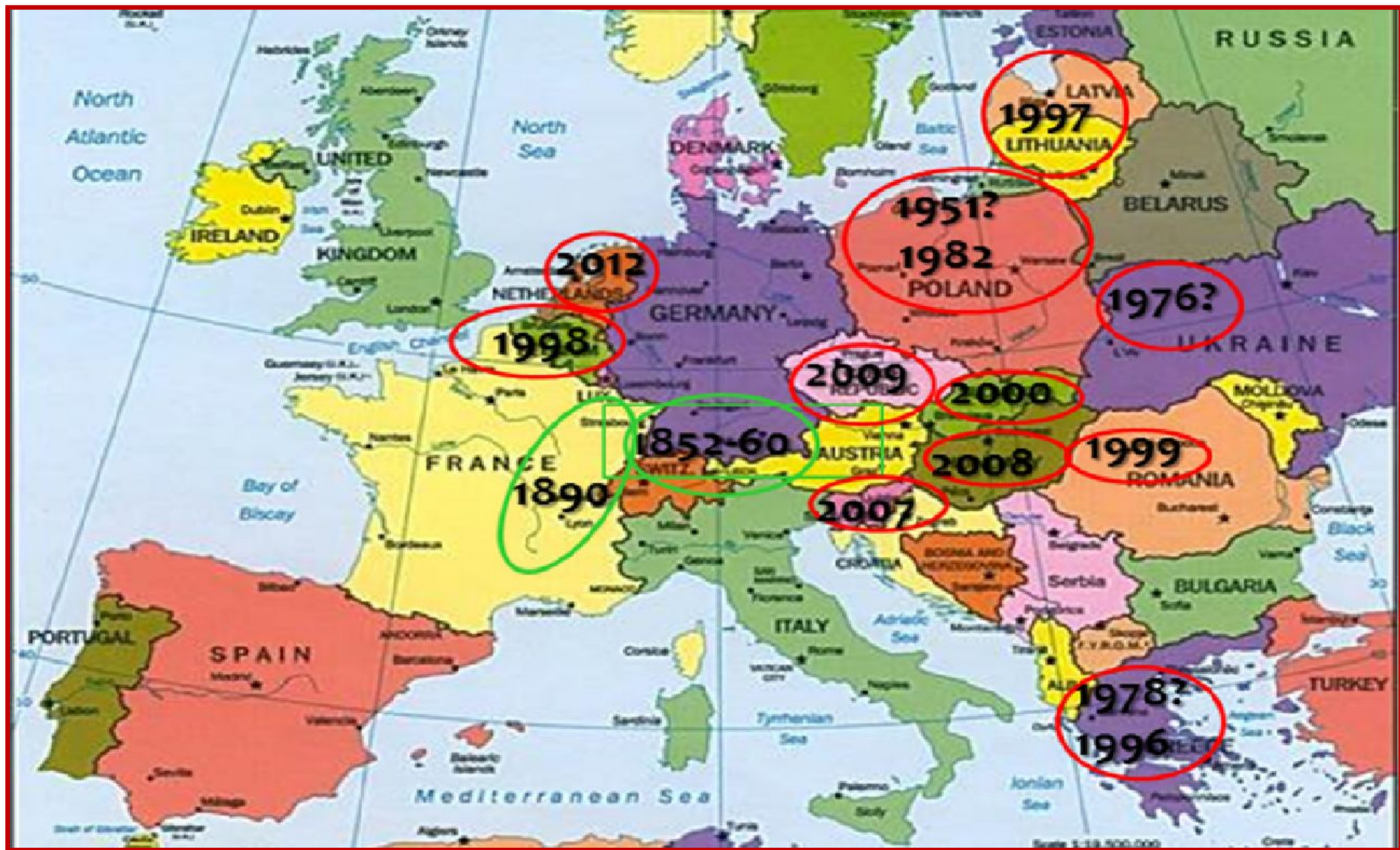
caused by infection with the larval stage of *E. multilocularis*

- *E. multilocularis*, a long tapeworm
- ~1-4 millimeter



Epidemiology

- AE is found across the globe ; especially prevalent in the northern latitudes of Europe, Asia & North America
- Earlier parasite of Northern Hemisphere
- Endemic area annual incidence: 0.03 to 1.2 per 100,000
- Mortality in untreated or inadequately treated: 90% within 10 to 15 years of diagnosis

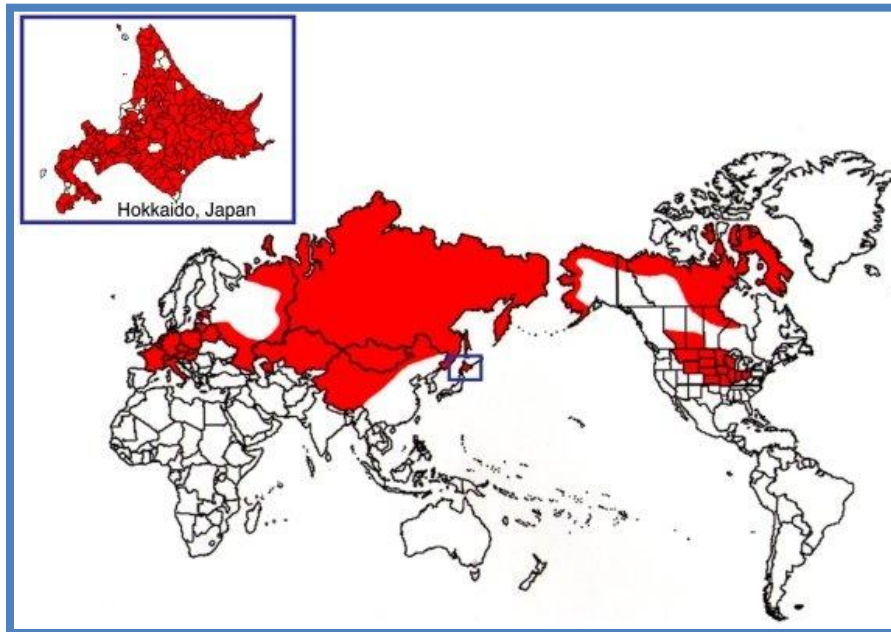


Historical map: Discovery of first cases of AE in various European countries

Green circles: historic endemic area; red circles: endemic areas discovered since the 1990s

Epidemiology

- AE is found worldwide, mostly in Northern latitudes
- Cases - Central Europe, Russia, China, Central Asia, Japan & North America



- Prevalence among wild foxes and coyotes can be high, and may reach over 50% in some areas; however, even in these areas, transmission to humans has been low

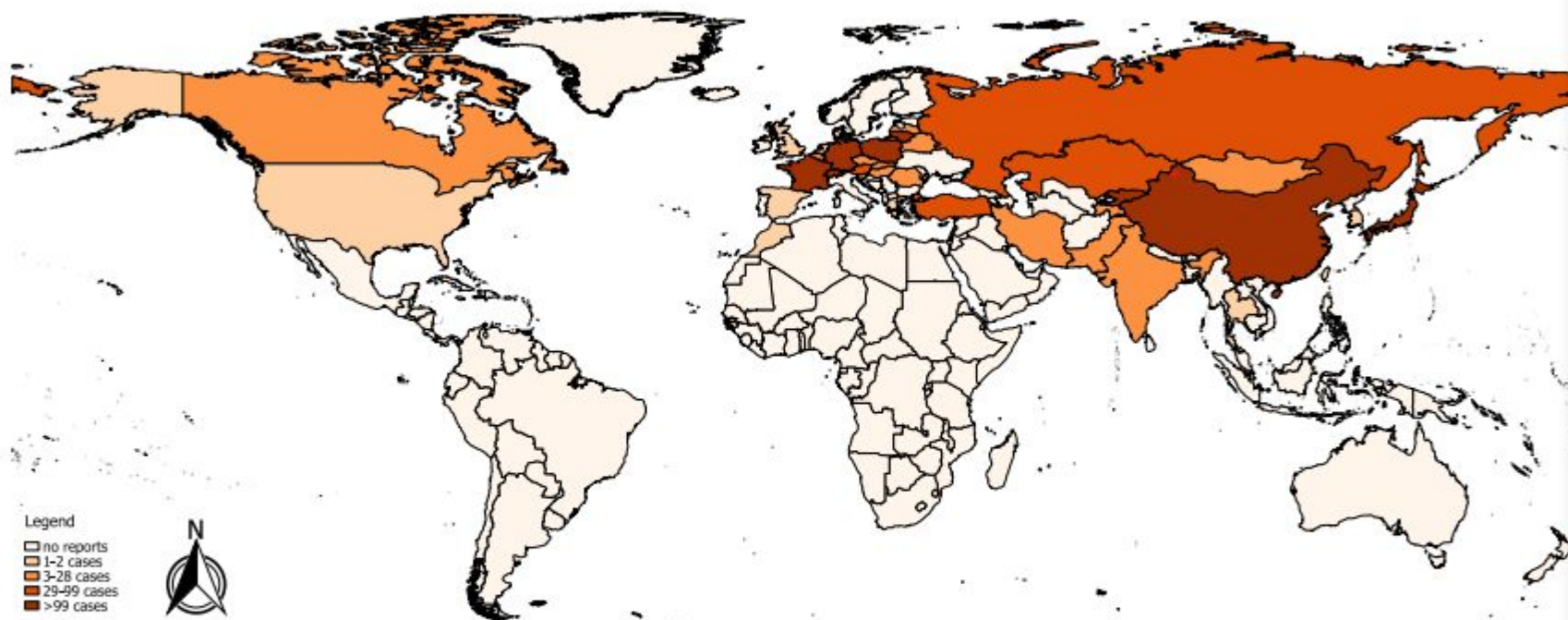
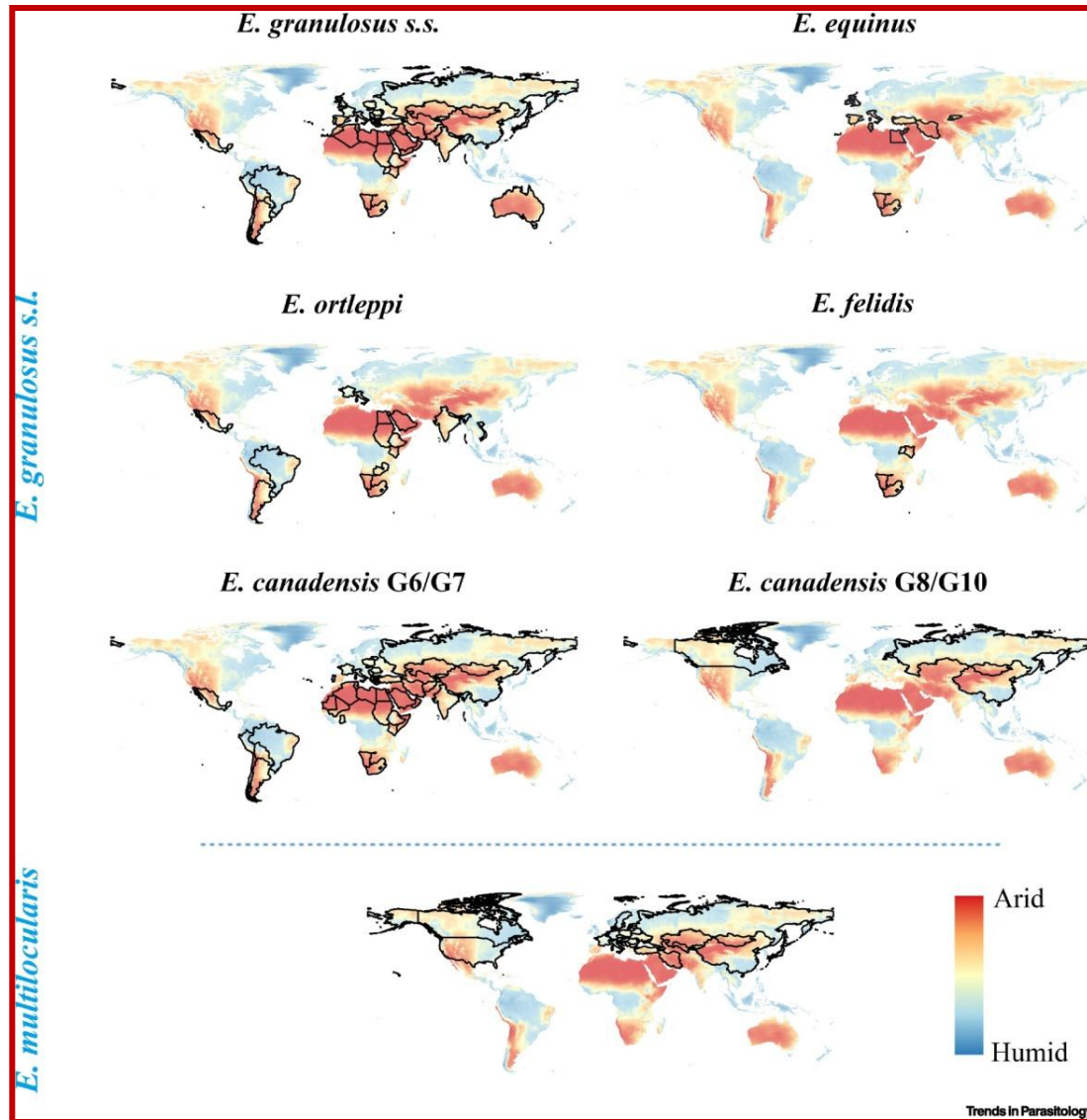


fig. 2 Worldwide distribution of alveolar echinococcosis in humans according to the published literature 2001–2018. Each country in which cases of AE had been reported in the literature between 2001 and 2018 was mapped. For the topographical colour shading of a country, the highest total number of cases in one reference within this period was the deciding factor



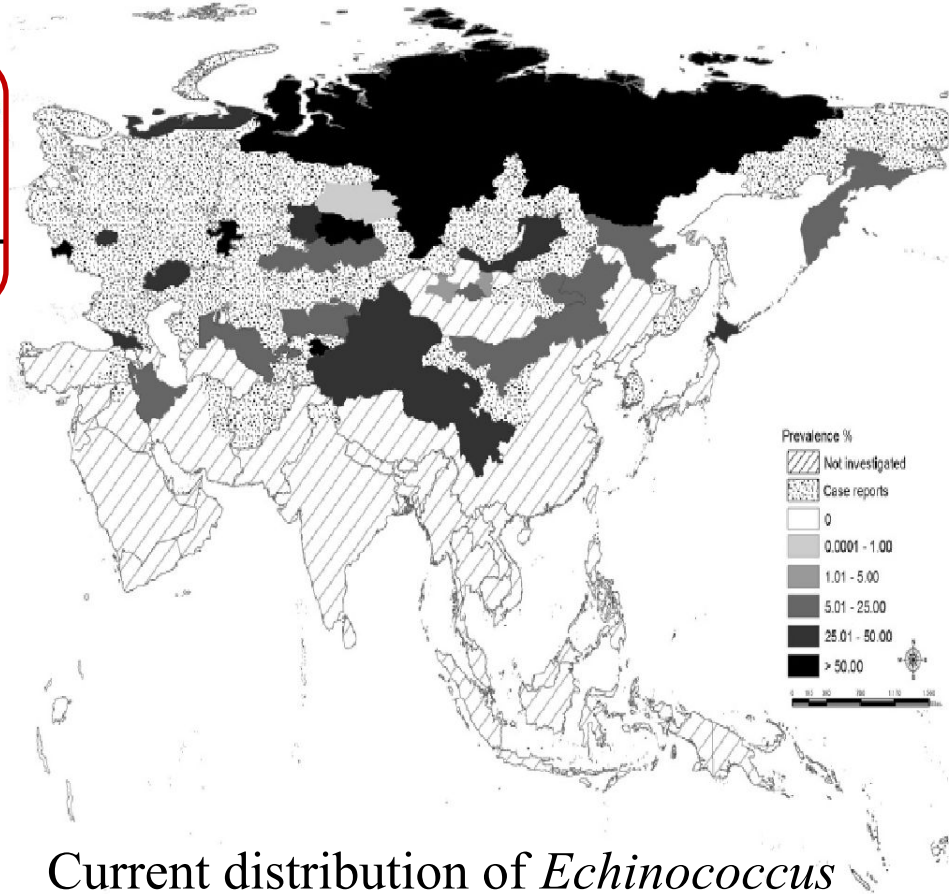
Epidemiology

- Public health concern in Northern Japan for past 40 years
- 91% of global cases from China
- China confirmed high incidence on Tibetan plateau

Global Distribution of Alveolar and Cystic Echinococcosis

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 M.F. Harandi^{||}, T. Romig[#], D. Antolova^{**}, J.M. Schurer^{§§, ¶¶},
 S. Lahmar^{|||}, G. Cringoli[§], J. Magambo^{##}, R.C.A. Thompson^{***},
 E.J. Jenkins^{§§}

Province	Population at risk (million)	Estimated prevalence	Estimated median number of new cases per year	Annual incidence per 10 ⁵ inhabitants
Gansu	3.6	2.9%	7676	2
Heilongjiang ^a			<10	0.02
Inner Mongolia	3.0	0.02%	44	0.2
Qinghai	5.4	1.0%	3766	67
Ningxia	1.2	2.0%	1770	2.7
Sichuan	0.92	3.6%	2390	2.9
Tibet	2.7	0.1%	172	5.5
Autonomous Region				
Xinjiang	5.8	0.2%	811	3.5



Current distribution of *Echinococcus multilocularis* in foxes in Asia

- In China, estimated number of AE cases per annum in various provinces
- 91% of global cases occur in China

Human Alveolar Echinococcosis after Fox Population Increase, Switzerland

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Johannes Eckert,* Bruno Gottstein,† Neriman Halkic,‡ Beat Muellhaupt,* Bettina Mareike Prinz,* Juerg
Reichen,† Philip E. Tarr,‡ Paul R. Torgerson,* and Peter Deplazes*

Table. Data from reported case-finding studies on human alveolar echinococcosis, 1956–2005, Switzerland

Study (reference no.)	Years	No. cases	Mean annual incidence/100,000 population	Mean age ±SD, y	Sex, no. (%)	
					Male	Female
Drolshammer (12)	1956–1969	122	0.15	54.2 ± 18.2	65 (53)*	57 (47)
Gloor (13)	1970–1983	145	0.16	55.0 ± 16.0	79 (54)*	66 (46)
Eckert (14)	1984–1992	71†	0.12	52.0 ± 17.7	33 (46)	38 (54)
This study	1993–2000	60	0.10	52.5 ± 18.4	26 (43)	34 (57)
This study	2001–2005	96	0.26‡	54.5 ± 17.3	42 (44)	54 (56)
Total	1956–2005	494	0.15	54.0 ± 17.3	245 (49.7)	248 (50.3)

- AE incidence doubled in previously recognized endemic areas of France, Switzerland, Germany, and Austria

Epidemiology of echinococcosis in Kazakhstan: an update

A. Abdybekova¹, A. Sultanov¹, B. Karatayev¹, A. Zhumabayeva¹,
Z. Shapiyeva², T. Yeshmuratov³, D. Toksanbayev³, R. Shalkeev⁴
and P.R. Torgerson^{5*}

- High endemicity of both *E. multilocularis* and *E. granulosus* in Kazakhstan and Kyrgyzstan
- Total 301 CE cases and mean age 33.8 years
- AE total 46 cases and mean age 35.6 years

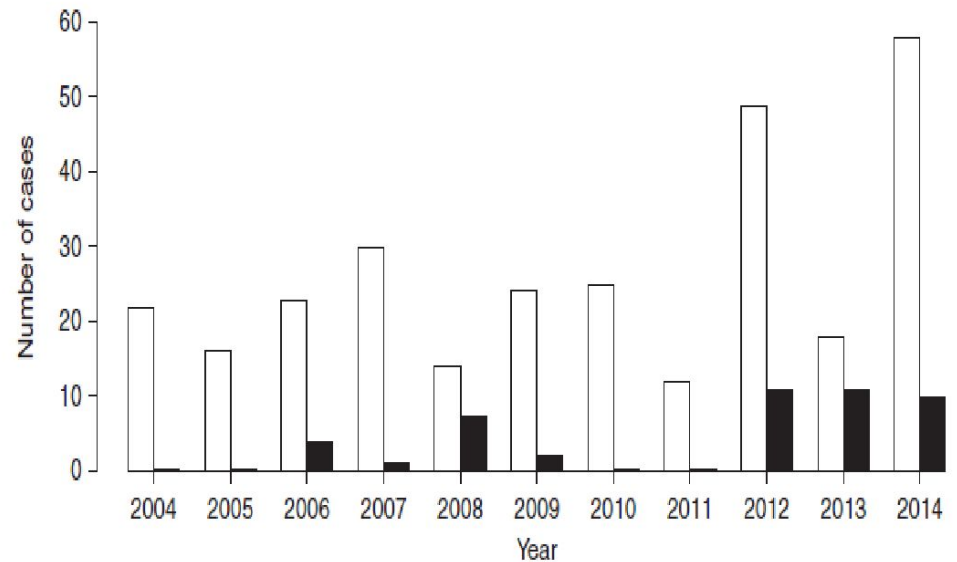


Fig. 2. Numbers of cases of cystic (CE; white bars) and alveolar (AE; black bars) echinococcosis cases treated in a single referral hospital in Almaty between 2004 and 2014.



Worldwide literature on epidemiology of human alveolar echinococcosis: a systematic review of research published in the twenty-first century

Sven Baumann¹ · Rong Shi² · Wenya Liu³ · Haihua Bao⁴ · Julian Schmidberger¹ · Wolfgang Kratzer¹ · Weixia Li⁴ · the interdisciplinary Echinococcosis Working Group Ulm

Country	Region	Total case number (<i>n</i>)	Prevalence (<i>n</i> /10 ⁵)	Incidence (<i>n</i> /10 ⁵ / year)	Period covered by data
India	Chandigarh ^e	1			n/a
		1			n/a
	Maharashtra	1			n/a
	Maharashtra	1			n/a
			1		n/a
			4 (4)		Mar 2010–May 2016
		3		n/a	



From 2001 to 2018

Life Cycle

- **Definitive hosts** – Dogs, Foxes, Wolves



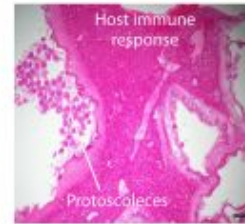
- **Intermediate hosts** - Small rodents and tundra voles



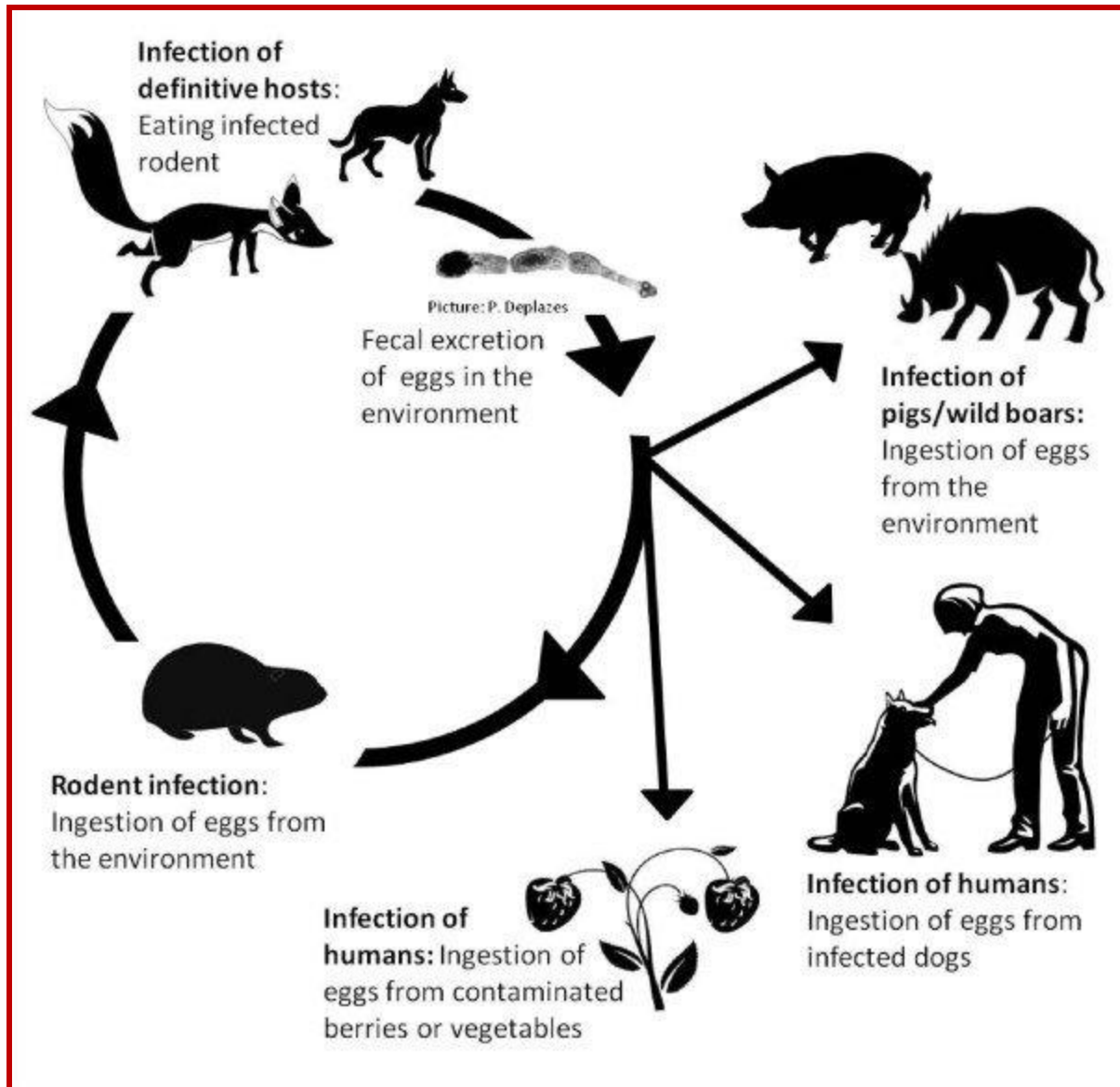
E. multilocularis



Dog or fox small intestine



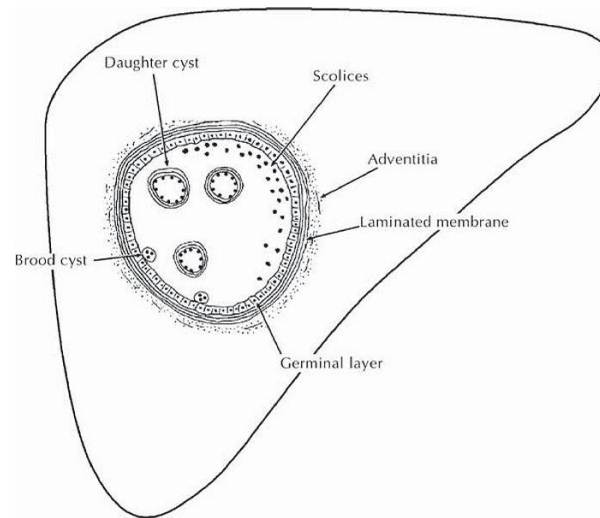
Liver of small mammals



Signs & Symptoms

- The primary infection of AE is in the liver, usually the right lobe
- Closely mimics hepatic carcinoma or cirrhosis and is more commonly diagnosed in people of an advanced age
- The larval mass resembles a malignancy in appearance & behavior
- Frequent ages at diagnosis usually between 35 & 65 years

- In chronic infections, the lesion consists of a central necrotic cavity filled with a white amorphous material that is covered with a thin peripheral layer of dense fibrous tissue
- Host tissue is directly invaded by extension of the budding and proliferating cyst wall, causing a pressure necrosis of surrounding host tissue
- Vigorous inflammatory & fibrous tissue reaction usually surrounds the larval mass



Hydatid Cyst of Liver

Main phases of infection

a. Initial phase:

- Always asymptomatic
- Cured spontaneously or turn to progressive course
- Incubation period: <5 to 15 years

b. Progressive phase:

- Metacestode infiltrated larger parts of liver or influences important functions
- Abdominal pain, jaundice, hepatomegaly, sometimes fever and anaemia, weight loss, and pleural pain

c. Advanced stage:

- Severe hepatic dysfunction, portal hypertension

d. Stable phase:

- In patients undergoing long-term chemotherapy
- Inhibit further parasite proliferation

e. Abortive phase:

- In asymptomatic patients: parasite dies out and mineralized

Neotropical Echinococcosis

- By larval stage of *E. vogeli* or *E. oligarthrus*
- Confined to Central and South America
- Around 250 cases till now

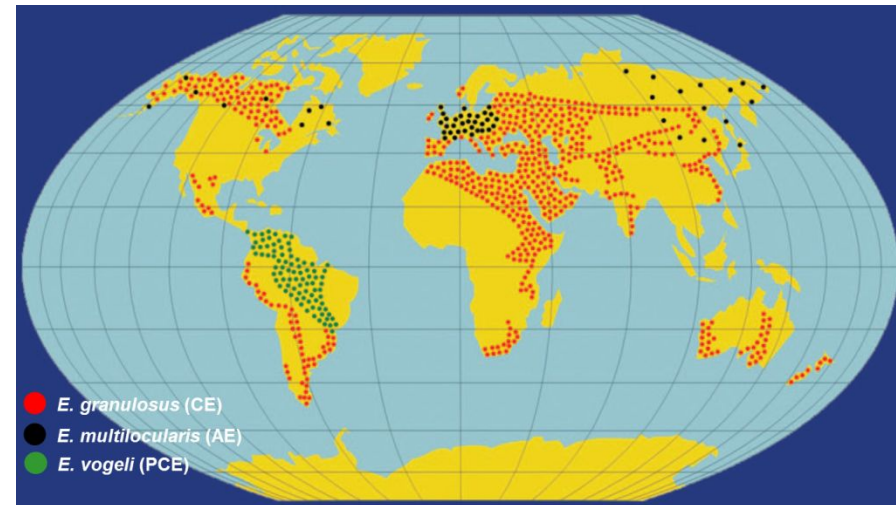
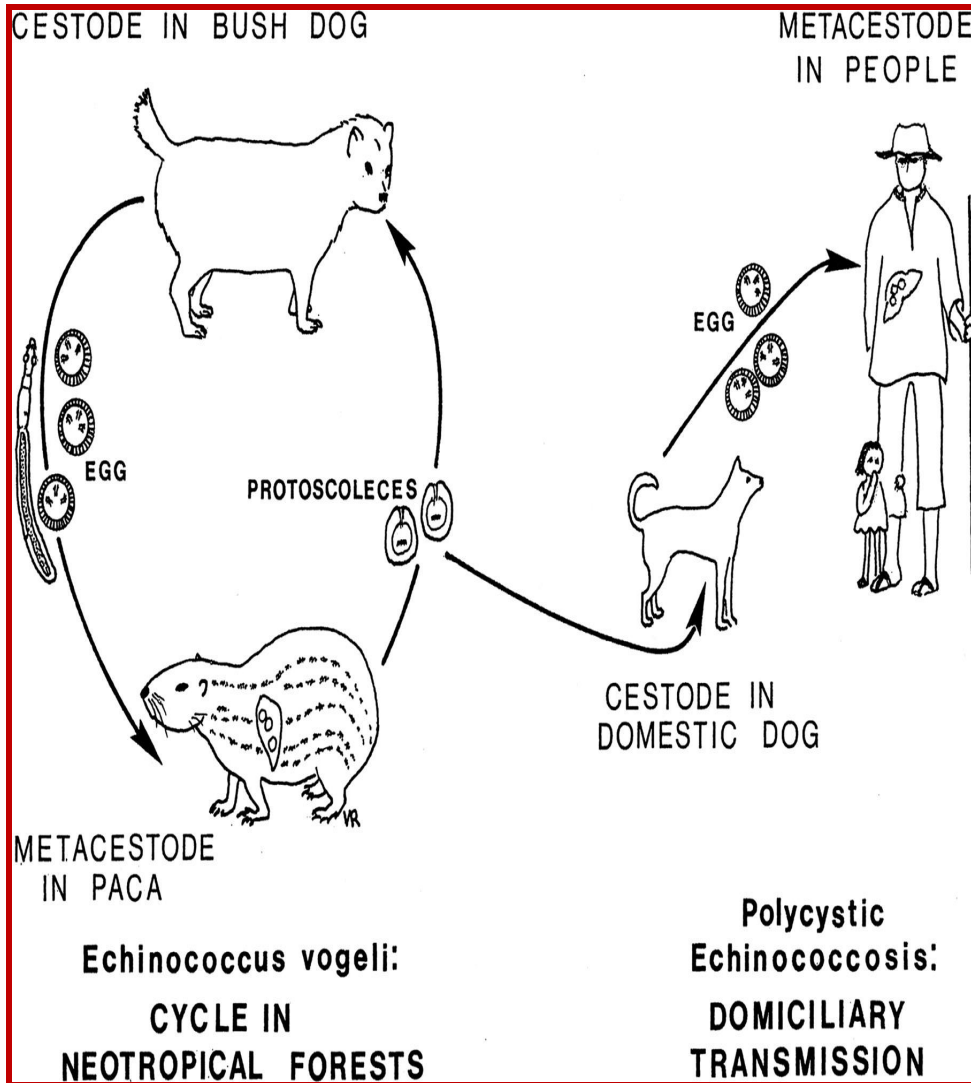


TABLE 4. Number of cases of echinococcosis in the Neotropics by country and species of *Echinococcus*^a

Country	No. of cases with indicated infection				Total
	<i>E. vogeli</i>	<i>E. cf. vogeli</i>	<i>E. oligarthrus</i>	<i>E. granulosus</i>	
Nicaragua		1			1
Costa Rica		1		1	2
Panama	2				2
Colombia	15	14			29
Ecuador	6	5			11
Venezuela	2	1	1		4
Peru	1				1
Brazil	21	77	1		99
Suriname	7	1	1		9
Uruguay		2			2
Argentina ^b		11			11
Chile		1			1
Total	54	114	3	1	172

^a As of March 2007. (Adapted and updated from reference 12 with permission from Elsevier.) *E. cf. vogeli*, *E. vogeli* causing infections with polycystic lesions but with no hooks found.

^b Synanthropic infections by *E. granulosus* not included.



<i>E. vogeli</i>	<i>E. oligarthrus</i>
Polycystic	Unicystic
Clinically more severe like AE	Less severe
Organ affected: liver(80%)>lungs> peritoneal cavity	Eyes & heart affected

First Report of *Echinococcus ortleppi* in Human Cases of Cystic Echinococcosis in Poland

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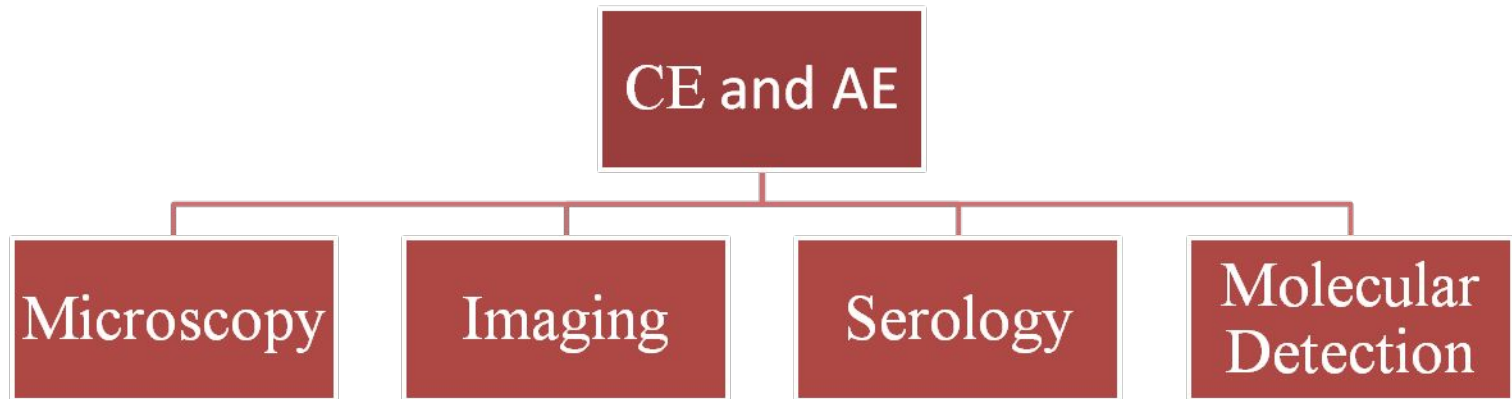
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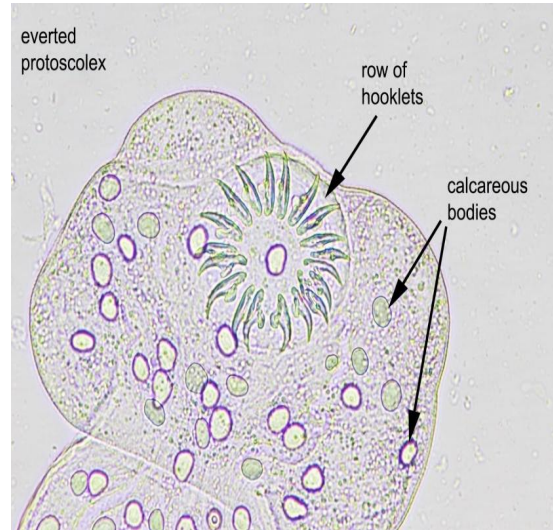
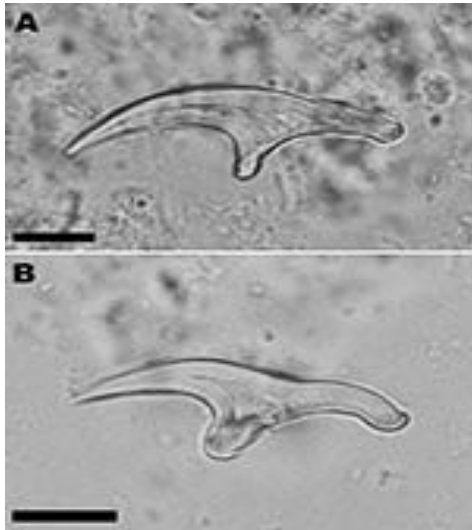
- Cyst fragment from 6 Polish female patients with diagnosed CE in year 2016-2017
- DNA extracted from the liver and lung samples
- 5 isolates: pig strain, *Echinococcus canadensis*
- One case 100% identical with *Echinococcus ortleppi*

DIAGNOSIS



Microscopy

- Hydatid fluid examined for scolex, hooklets or fragments of laminated membrane
- Wet mount examination



Imaging

- Essential for diagnosis

X-ray	Ultrasound (US)	Contrast-enhanced ultrasonography (CEUS):	CT scan & MRI
Lung cysts	Method of choice Widely used to diagnose CE or AE liver lesions	Detect small cysts (<2 cm in diameter)	An important preoperative diagnostic tool to determine vascular, biliary or extra hepatic extension. In CE, MRI better diagnostic value than CT scan
	Best method for early diagnosis in endemic area for mass screening	Differentiate AE from abscesses and tumors	Both procedures complementary for AE

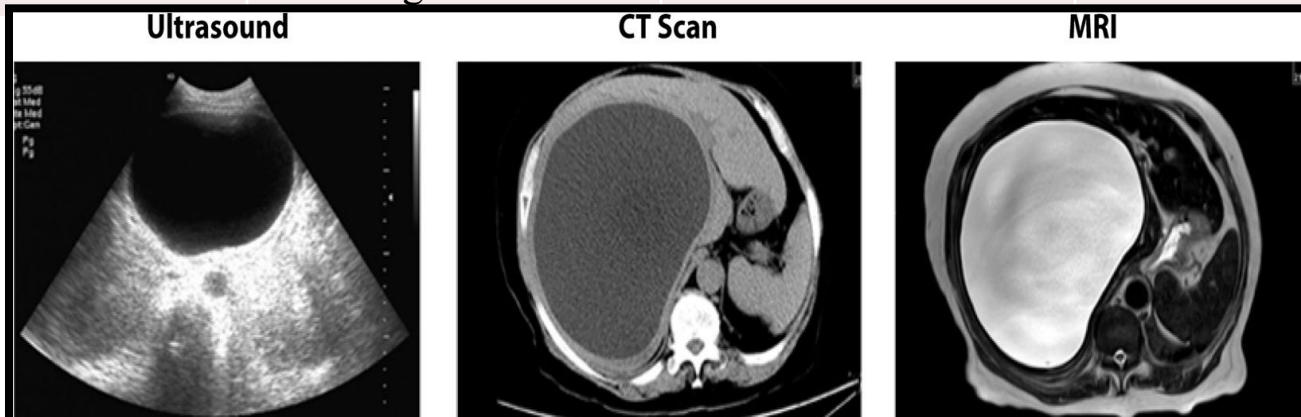


Table 1: Sonographic classification of hydatid cysts

Gharbi Type	WHO Type	Cyst Morphology
I	CE 1	Unilocular anechoic lesion with double line sign
III	CE 2	Multiseptated rosette like honeycomb cyst
II	CE 3A	Cyst with detached membranes (water-lily sign)
III	CE 3B	Cyst with daughter cysts in solid matrix
IV	CE 4	Cyst with heterogenous hypoechoic/hyperechoic contents. No daughter cysts
V	CE 5	Solid plus calcified wall

TABLE 2 WHO Informal Working Group on Echinococcosis PNM classification and staging of alveolar echinococcosis

Classification	Description
P	Hepatic localization of the parasite
PX	Primary AE lesion cannot be assessed
P0	No detectable AE lesion in the liver
P1	Peripheral lesion(s) without proximal vascular and/or biliary involvement
P2	Central AE lesion(s) with proximal vascular and/or biliary involvement of one lobe ^b
P3	Central lesion(s) with hilum vascular or biliary involvement of both lobes and/or with involvement
P4	Any liver lesion with extension along the vessels ^c and the biliary tree
N	Extrahepatic involvement of neighboring organs (diaphragm, lung, pleura, pericardium, heart, gastric glands, adrenal glands, peritoneum, retroperitoneum, parietal wall [muscles, skin, bone], pancreas, regional liver ligaments, kidney)
NX	Not evaluable
N0	No regional involvement ^d
N1	Regional involvement of contiguous organs or tissues
M	Absence or presence of distant metastases (lung, distant lymph nodes, spleen, central nervous system, muscle, kidney, distant peritoneum, and retroperitoneum)
MX	Not completely evaluated
M0	No metastasis ^e
M1	Metastasis
PNM stages	
I	P1 N0 M0
II	P2 N0 M0
IIIa	P3 N0 M0
IIIb	P1-P3 N1 M0, P4 N0 M0
IV	P4 N1 M0, any P any N and/or M1

Serology



Serologic Tests

i. Enzyme immunoassay/enzyme linked immunosorbent assay

- ✓ Detection of IgG antibodies implies exposure to the parasite, while in active infection high titers of specific IgM and IgA antibodies are observed
- ✓ Detection of circulating hydatid antigen in the serum is of use in monitoring after surgery and pharmacotherapy and in prognosis
- Liver cysts are more **likely to** yield positive serologic test results than pulmonary cysts
- Positive test results are less **likely with** calcified or dead cysts and more likely with ruptured cysts

For CE

Antigens	Source	Use
Lipoproteins antigen B (AgB)	Hydatid fluid	Widely used in serological assays for CE
Lipoproteins antigen 5		
Enriched antigen 5	<ul style="list-style-type: none">• Novel antigens• >90% sensitivity and specificity on selected serum samples	Never been evaluated on large scale
Recombinant antigens based on repeated tandem E. granulosus AgB (2B2t antigen)		
Recombinant Ag5		
Tegumental protein EgTeg		
Alkaline phosphatase (EgAP)		
EpC1		

For AE

Antigen (excretory/secretory (ES) fraction of intact metacestodes)		
Em2	Specific for <i>E. multilocularis</i> infection	<ul style="list-style-type: none">• Commercialized Em2plus ELISA• Used extensively• Sensitivity and specificity >90%
Em492		
EmAP		
EmP2		
Em10		Show high diagnostic performance for confirming AE
EmII/3	Derivative of Em10	
Em18		

Echinococcosis form and test	Antigen ^b	Sensitivity (%)	Relative specificity ^c (%)	Cross-reactions
Cystic				
IgG ELISA	Crude EgCF	80–>99	61.7	Cestodes (89%), trematodes (30%) nematodes (39%)
	Antigen B (native or synthetic peptide)	63–92	85–93	AE
IgG4 ELISA	Crude EgCF	61–67	>99	AE only (see AE)
EITB ^d	Crude EgCF	71	>98 ^e	<i>T. solium</i> cysticercosis only
	Antigen B fraction	92	100	None
	Antigen B subunits	34–36	>90	
Alveolar				
IgG ELISA	Crude EgCF	97.1	61.7	See above
	Em2PLUS	97.1	98.9	CE (25%)
	Em2/Em2G11	89.3	100	CE (5.6%)
	Em II/3–10	86.4	98.4	CE (6.5%)
IgG4 ELISA	Crude EgCF	48–67	>99	CE (see CE)
EITB	Em18	97	100	None
	Glycoproteins	70–90	>95	

Tests for antibody detection in human CE & AE

Immunodiagnosis of cystic echinococcosis by antigen detection in serum, urine, and saliva samples

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Department of Parasitology, Postgraduate Institute of Medical Education and Research, Chandigarh, India

Table 1: Hydatid antigen detection by ELISA in patients and controls

	Subjects tested	Number positive		
		Serum ^a	Saliva ^b	Urine ^c
Hydatidosis patients				
i) Clinically and radiologically diagnosed	21	8	5	11
ii) Surgically confirmed	4	2	1	2
Total (i + ii) [#]	25	10	6	13
iii) Clinically suspected	25	8	3	5
Controls (N = 40)				
Cysticercosis	5	2	2	4
Ascariasis	5	1	2	3
Amebic liver abscess	5	0	1	1
Healthy controls	25	0	0	0
Sensitivity* (%)		40	24	52
Specificity (%)		92.5	87.5	80
Positive predictive value (%)		76.9	54.5	61.9
Negative predictive value (%)		71.1	64.8	72.7

Molecular Methods

```
graph TD; A[Molecular Methods] --> B[Quantitative PCR  
Nested PCR]; A --> C[Loop-Mediated Isothermal Amplification(LAMP)]
```

Quantitative PCR Nested PCR

- Distinguish *Echinococcus* species
- Discriminate genotypes of *E. granulosus*
- Used on biopsy or fine-needle cytology specimens for AE diagnosis with unusual imaging aspects and / or with negative serology

Loop-Mediated Isothermal Amplification(LAMP)

- Can be used as first-line screen for *Echinococcus* spp. in field
- Can detect *Echinococcus* sp. egg DNA in environmental samples

Rapid Diagnostic Tests

Publication	RDT format	Antigen used	Production
Tamarozzi et al. (2016) [22]	ICT	AgB/Ag5	Commercial: VIRapid HYDATIDOSIS (Vircell, Spain)
	ICT	rAgB	Commercial: ADAMU-CE (ICST, Japan)
	DIGFA	HCF/Psc/AgB/Em2	Commercial: Echinococcus DIGFA (Unibiotest, China)
Xie et al. (2015) [in Chinese] [27]*	ICT (read with a reader)	N/S	In house
Chen et al. (2015) [6]	DIGFA	rAgB	In house
Santivanez et al. (2015) [18]	ICT	rAgB	Commercial: ADAMU-CE (ICST, Japan) different version from test used by Tamarozzi et al.
Tamer et al. Med Sci Monit (2015) [23]	ICT	AgB/Ag5	Commercial: VIRapid HYDATIDOSIS (Vircell, Spain)
Wang et al. Parasitol Res (2013) [25]	ICT	HCF	In house
Feng et al. Acta Trop (2010) [8]	DIGFA	HCF/Psc/AgB/Em2	Commercial: Echinococcus DIGFA (Unibiotest, China)
Olut et al. (2005) [15]	Dot-immunobinding	HCF	Echinostrip, developed by Lofarma Laboratories, Italy
Al-Sherbiny et al. (2004) [1]	Dipstick	HCF	In house
Babba et al. (2004) [2]	Slide-LAT ^a	HCF	In house
Barbieri et al. (1993) [3]	Slide-LAT	HCF lipoprotein fraction	In house
Rogan et al. (1991) [17]	Rapid dot-ELISA	AgB	In house
Hoghooghi et al. (1976) [11]	Slide-LAT	N/S	Commercial: Italdiagnostico Co, Italy (no more on the market)
Varela-Diaz et al. (1975) [24]	Slide-LAT ^a	HCF	In house
Kagan et al. (1966) [12]	Slide-LAT ^a	HCF	In house
Szyfres and Kagan (1963) [21]	Slide-LAT	HCF	In house

Comparison of the Diagnostic Accuracy of Three Rapid Tests for the Serodiagnosis of Hepatic Cystic Echinococcosis in Humans

Francesca Tamarozzi^{1,2*}, Ilaria Covini¹, Mara Mariconti^{1,2}, Roberta Narra^{1,2}, Carmine Tinelli³, Annalisa De Silvestri³, Federica Manzoni³, Adriano Casulli⁴, Akira Ito⁵, Andreas Neumayr⁶, Enrico Brunetti^{1,2,7}

Test	Sensitivity	Specificity
VIRapid	74.1% (61.0–84.7)	96% (79.6– 99.9)
DIGFA	72.9% (59.7–83.6)	72% (50.6– 87.9)
ADAMU-CE	57.6% (44.1–70.4)	100% (86.3– 100)
ELISA (reference test)	69.5% (56.1–80.8)	96% (79.6– 99.9)

Casoni Reaction

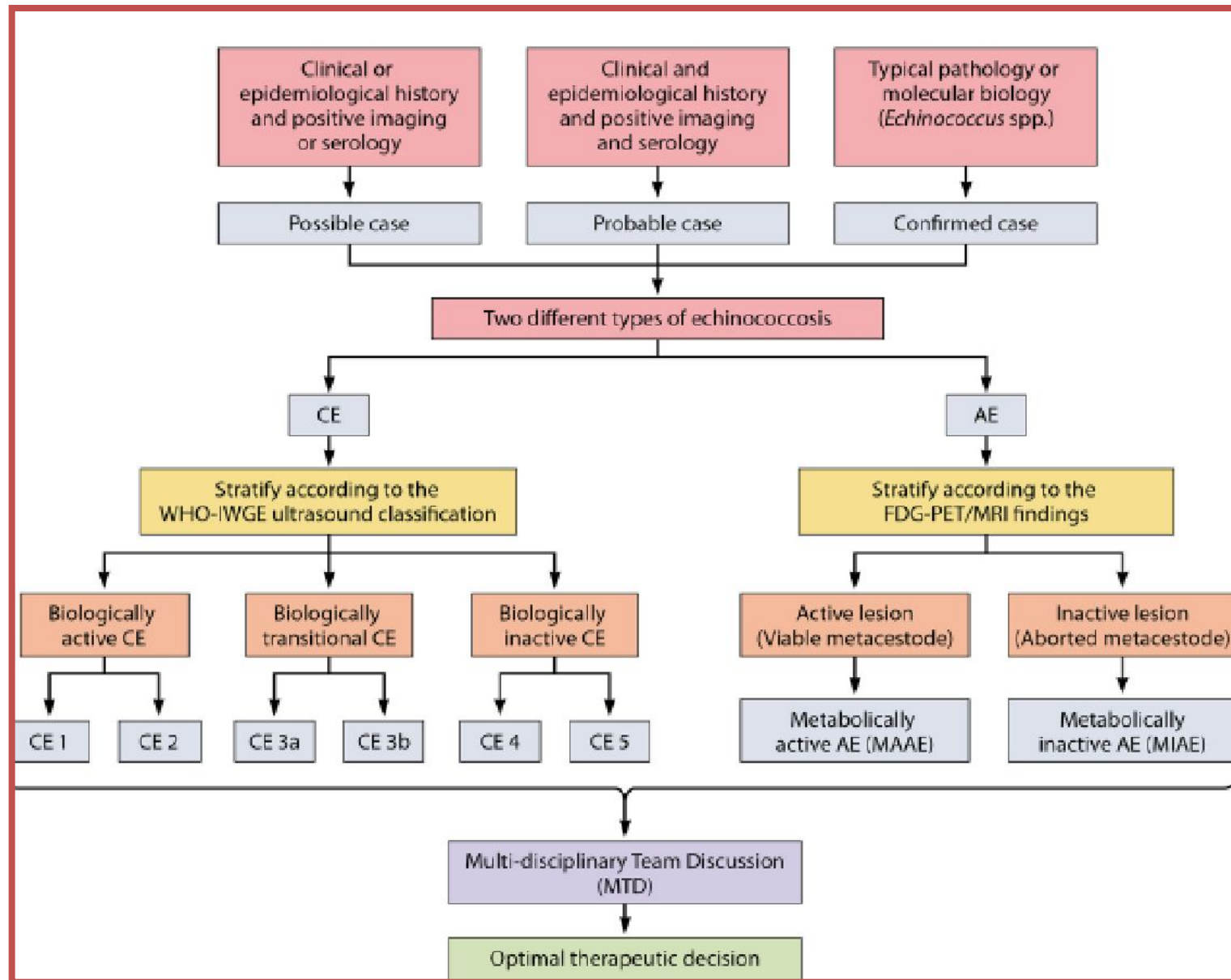
- An immediate hypersensitivity skin test introduced by Casoni in 1911
- Reagent is pooled hydatid fluid containing scolices being usually selected
- Skin of the outer half of the upper arm is most suitable for the test
- Fluid is injected intradermally in amount sufficient in fresh area of skin to raise a white area approx one cm in diameter
- Usually about 0.2 ml of the fluid is required
- Sterile normal saline, 0.2 ml is injected in other arm for control



- Control wheal fades rapidly, while that from the hydatid fluid increases
- A zone of erythema surrounds the wheal, which reaches its maximal dimensions within half an hour and then fades quickly
- Reactions are recorded as positive when the wheal measures at least 2-4 cms



The typical skin reaction in a Positive Casoni Test



Algorithm for diagnosis of CE and AE

Treatment & Management

Four treatment approaches:

1. Anti-infective drug treatment

Albendazole is the drug of choice because of its greater absorption from the GI tract to achieve higher plasma levels

2. Surgery

- Successful in simple cyst
- Impractical in multiple cyst, high surgical risk



The cyst appears creamy and smooth

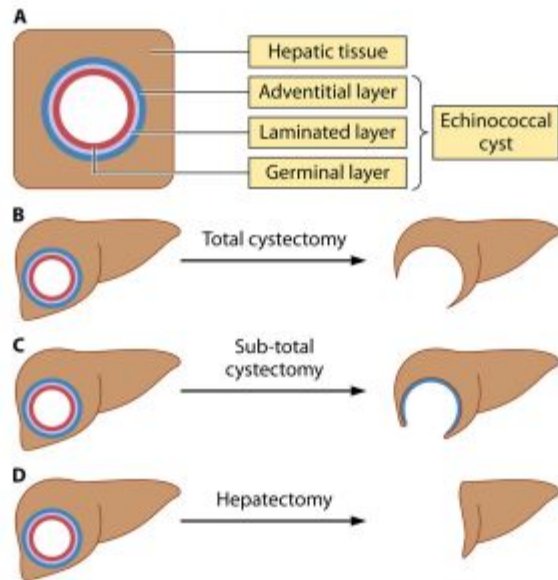
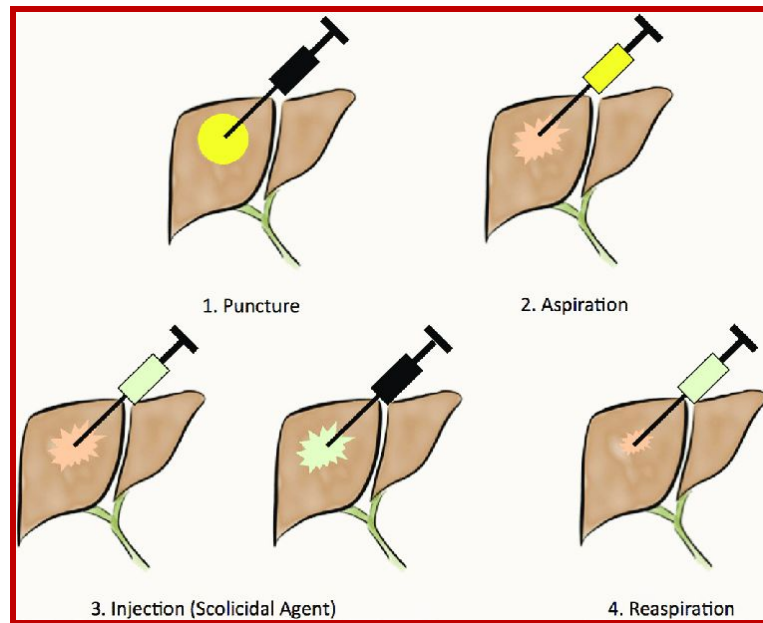


FIG 8 Schematic structure of the echinococcal cyst and different approaches for surgical removal. (A) The echinococcal cyst is made up of the adventitial layer, laminated layer, and germinal layer (from outside to inside). (B) Total cystectomy involves resection of the entire adventitial layer ("subadventitial resection"), the laminated layer, and the germinal layer. (C) Sub-total cystectomy involves partial resection of the adventitial layer and total resection of the laminated layer and the germinal layer, leaving parts of the adventitial layer in place whenever the operation is difficult because of the proximity of large vessels and/or adhesions. (D) Hepatectomy involves the *en bloc* resection of the echinococcal cyst along with part of the normal liver parenchyma. Partial cystectomy, which requires opening of the cyst, may leave all or part of the laminated layer and germinal layer and relies on the efficacy of a protoscolecide to destroy the metacestode; it should generally not be considered because of the potential for recurrence.

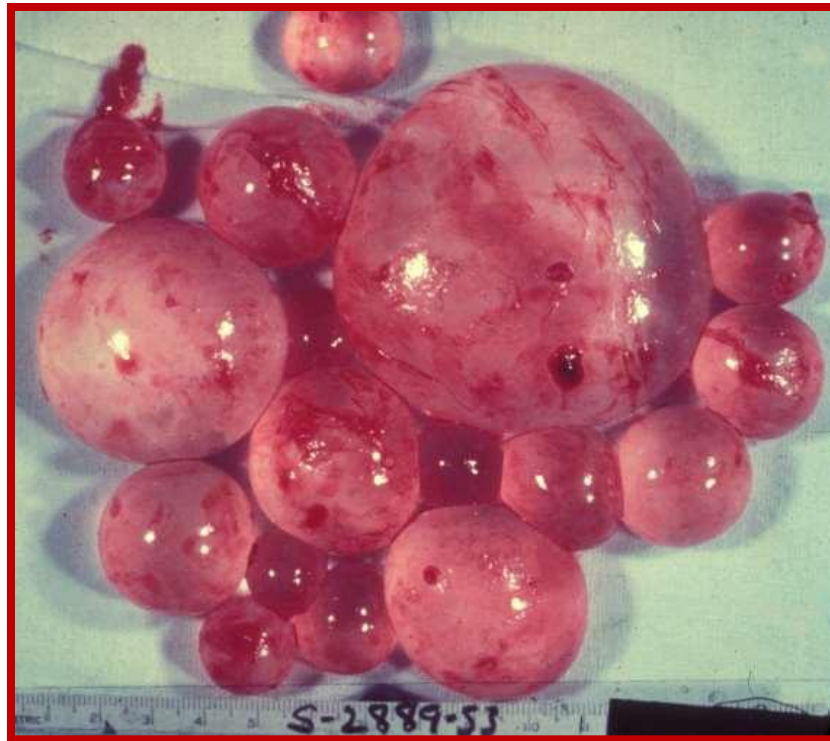
3. Percutaneous treatment of the hydatid cysts with the **PAIR** (Puncture , Aspiration, Injection, Re-aspiration) technique

- Minimal invasive technique
- Aspiration of cyst fluid → injection of parasitocidal solution (95%ethanol or 15%NaCl solution) → Reaspiration



4. “Watch and wait”

Hydatid cysts



Excised hydatid cysts showing infection with *Echinococcus*

“Watch and wait”

- Idea of leaving certain cyst types untreated and just monitoring them over time is a logical consequence of two main findings:
 - 1) a good proportion of cysts are consolidating and calcifying (i.e., become completely inactive) without any treatment
 - 2) cysts that have arrived at this stage and behave quietly (i.e., do not compromise organ functions or cause discomfort) seem to remain like this or stabilize even further
 - 3) This decision must, however, be accompanied and verified by long-term ultrasonographic follow-up for a period up to ten years

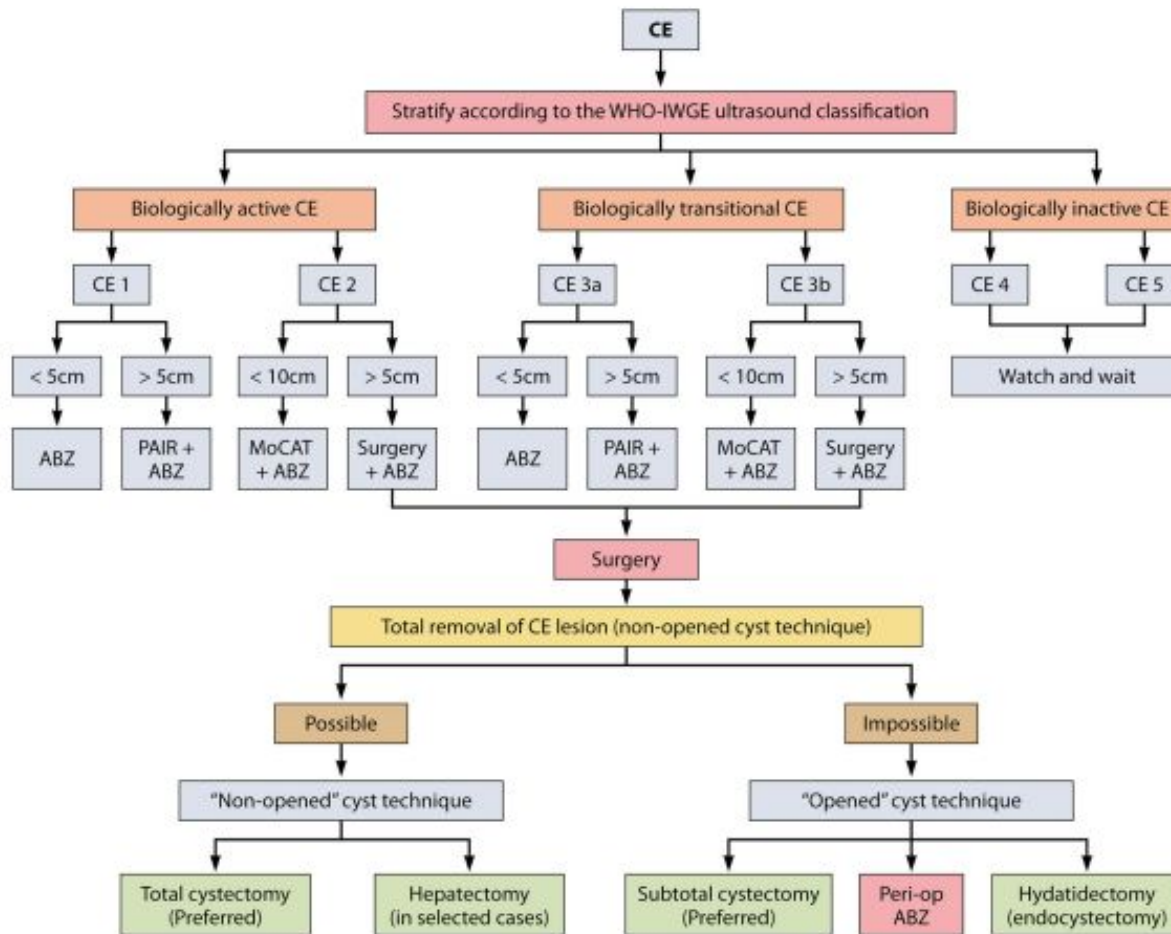


FIG 7 Algorithm for the treatment of cystic echinococcosis (CE), based on "WHO-IWGE international classification of ultrasound images in cystic echinococcosis for application in clinical and field epidemiological settings" (86) and Fig. 4 and 5. WHO-IWGE, World Health Organization Informal Working Group on Echinococcosis; ABZ, albendazole; PAIR, puncture-aspiration-injection-reaspiration (nonsurgical percutaneous interventional technique for treatment of CE cysts).

Algorithm for treatment of CE

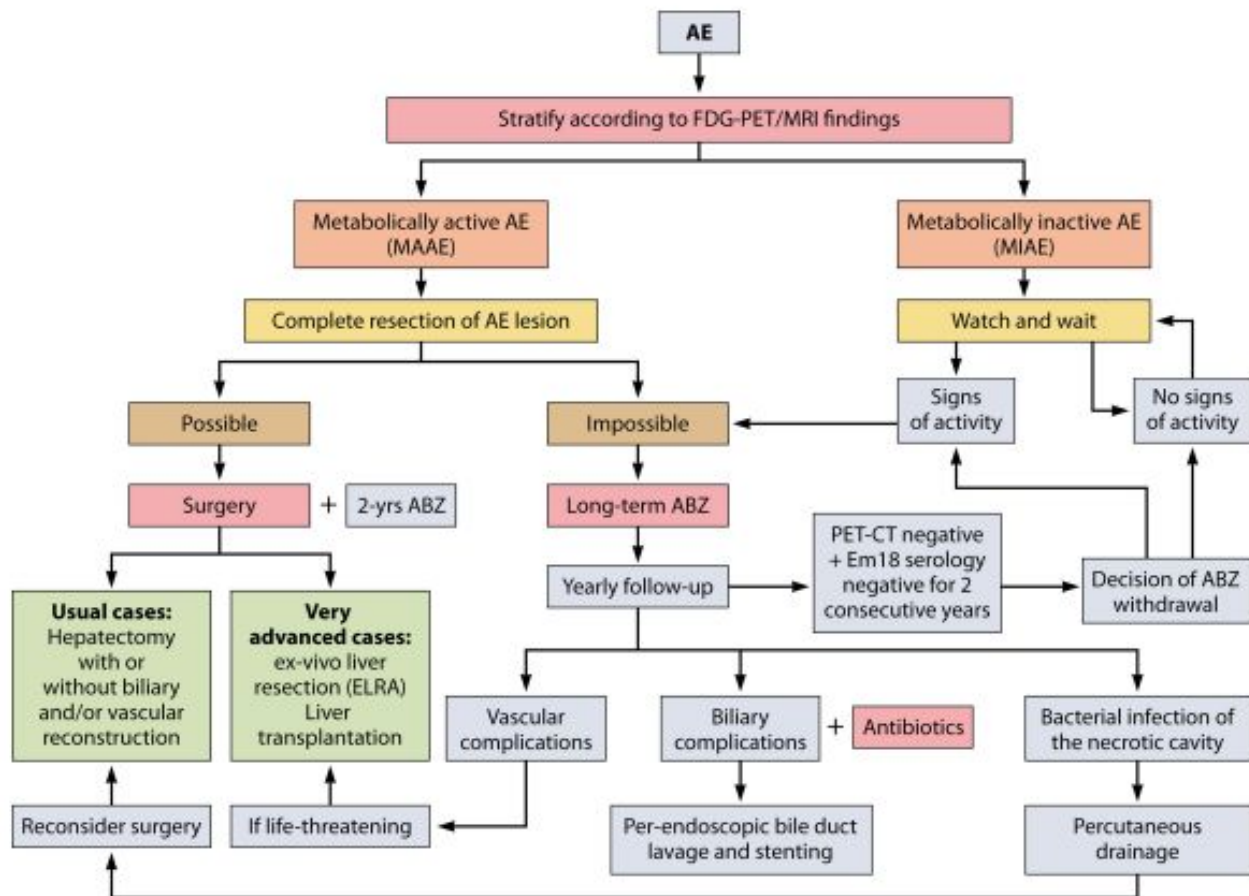


FIG 9 Algorithm for the treatment of alveolar echinococcosis. FDG-PET, fluorodeoxyglucose-positron emission tomography (increased uptake of FDG by the periparasitic immune response is the currently accepted evidence for AE lesion metabolic activity) (94). MRI, magnetic resonance imaging (identification of typical microcysts on T2-weighted images at MRI is a surrogate marker for AE lesion metabolic activity) (98). ABZ, albendazole; ELRA, ex vivo liver resection with autotransplantation.

Algorithm for treatment of AE

Prognosis

- Prognosis is generally good
- Complete cure is possible with total surgical excision without spillage
- Spillage occurs in 2-25% of cases
- Operative mortality rate varies from 0.5-4%

Control and Prevention for CE

Control of Definitive host

Type I:

- Long-term measures of public health education
- Improvement of slaughter hygiene and meat inspection
- Dog registration
- Sanitation measures

Type II:

- Based on legislation
- Measures to interrupt parasite transmission:
 - Stray dog control
 - Registration of all owned dogs
 - Spaying of bitches
 - Control measures in dogs and sheep, as part of a One-Health approach include

Deworming of dogs with praziquantel at least 4 times / year

Vaccination of lambs with EG95 vaccine

Prevention

- Controlled by preventing transmission of the parasite
- Prevention measures include limiting the areas where dogs are allowed
- Prevent dogs from feeding on the meat of infected sheep
- Control stray dog populations
- Restrict home slaughter of sheep and other livestock
- Do not consume any food or water contaminated by fecal matter from dogs
- Wash your hands



Control And Prevention for AE

Control in definitive host:

- Eliminating *E. multilocularis* from its wild animal hosts is next to impossible; therefore, contact with dogs and foxes in areas where the infection is endemic should be avoided
- Do not allow dogs to feed on rodents and other wild animals
- Avoid contact with wild animals such as foxes, coyotes & stray dogs
- Regular deworming of domestic carnivores that have access to wild rodents
- Culling of foxes and unowned free-roaming dogs appears to be highly inefficient

Precautions for individuals:

- Persons at special risk: repeated serological screening
- Special safety precautions for laboratory and field workers
- Do not encourage wild animals to come close to your home or keep them as pets
- Wash your hands

Measures to reduce morbidity and mortality:

- Detecting human AE cases at an early stage
- Stepwise introduction of regular screening programs for areas with relatively high incidence rates of human AE or high prevalence rates of parasite in definitive hosts

Surveillance:

- Adequate surveillance: detect or exclude AE spread and emergence
- Mass screening of living foxes, dogs, and cats by ELISA
- Detect parasite DNA in fecal samples by PCR: confirmation test in selected case

TRANSACTIONS OF THE ROYAL SOCIETY OF TROPICAL MEDICINE AND HYGIENE, VOL. 72, No. 6, 1978

Echinococcus multilocularis infection in India: first case report proved at autopsy

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Summary

The occurrence of *Echinococcus multilocularis* is reported in India for the first time. The patient was a young man, various clinical diagnoses were made and he finally died after an attempted membranotomy for suspected membranous obstruction in the inferior vena cava. Autopsy revealed classical *E. multilocularis* infection of the liver with direct spread of the inferior vena cava, the right atrium and through the diaphragm into the base of the left lung. It also had caused an outflow tract obstruction to the hepatic venous flow by direct physical pressure distorting the proximal intrahepatic portion of the inferior vena cava. In addition the patient had multi-valvular lesions of rheumatic origin and a terminal infective endocarditis due to staphylococcal infection.

Introduction

A recent review (MATOSSIAN *et al.*, 1977) emphasizes the global nature of the problem of hydatidosis. Reports of hydatid disease from India are many (MAPLESTONE, 1933; SAMI, 1938; REDDY & SUVARNA KUMARI, 1971; REDDY *et al.*, 1968; PRAKASH *et al.*, 1967) but all have recorded *E. granulosus*. *E. granulosus* is known to involve the portal vein giving rise to portal hypertension (DALAL, GUPTA & JOSHI, 1977). *E. multilocularis* has been reported mainly from Europe, Iran, Japan, America and the Soviet Union, particularly Siberia and the Central Asian Republics and there have been about 52 cases recorded from Turkey (YASAROL, 1974). *E. multilocularis* infection in man causes alveolar hydatid which behaves like a tumour and rapidly progresses with metastases to various organs. Its life-cycle involves wild carnivores (e.g. foxes and wolves) and dogs as final hosts and field mice and tundra voles as intermediate hosts. Man becomes infected accidentally by eating wild berries etc. contaminated with fox, wolf or dog faeces. The morphology is very characteristic. No case has yet been reported from the Indian subcontinent and this we believe is the first case of its kind from India.

Case Report

B.A.K., a 29-year-old male was first a patient in the All India Institute of Medical Sciences, New Delhi between 23rd February and 7th March, 1977 with the diagnosis of hepatosplenomegaly (? tuberculous), non-specific reactive hepatitis, as evidenced

by a liver biopsy, and anxiety neurosis. At that time his liver was enlarged 4 cm below the costal margin and was tender. The spleen was 3 cm below the costal margin and firm. He was discharged on anti-tuberculous treatment on a presumptive diagnosis of tuberculosis. On 27th July, he was admitted to the Postgraduate Institute of Medical Education and Research, Chandigarh (C.R. No. 333531) with complaints of chest pain and dyspnoea on exertion for the last one and a half years. He had had one bout of haematemesis 18 months previously. Physical examination showed marked pallor, oedema of the feet, B.P. 110/60, JVP 6 cm and firm. His family and personal histories contributed nothing. Various clinical conditions were considered, e.g. non-cirrhotic portal fibrosis, amoebic liver abscess, angina pectoris with congestive cardiac failure, portal hypertension with cirrhosis and hydatid cyst of the liver. Cardiology consultation revealed rheumatic heart disease with mitral stenosis, tricuspid stenosis, tricuspid incompetence and aortic incompetence.

On 11th August, a Casoni test was done which gave an immediate positive reaction. Some observers considered that the feel of the liver was different from that of an hydatid cyst and suggested the possibilities of gumma or hepatoma.

On 14th September, catheter studies revealed a filling defect of the hepatic veins and a block in the inferior vena cava in the area where it passes under the liver. The upper end of the block could not be clearly defined. A liver scan showed filling defect in the postero-superior portion.

On 5th October, the patient developed jaundice. A liver biopsy done on 22nd October showed non-specific changes.

On 26th November, the case was reviewed and a membrane causing an inferior vena caval block leading to cirrhosis and hepatoma was strongly suspected.

On 1st December, an exploratory thoracotomy was undertaken with a view to performing a finger fracture membranotomy. On opening the pericardium, the inferior vena cava was observed to be solid with a mass. Its opening into the right atrium could not be negotiated. On opening

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the diaphragm about 200 ml of frank 'pus' was seen in the sub-diaphragmatic region. There was a large, necrotic, greyish mass in the postero-superior aspect of the liver, which was interpreted as a malignant tumour. Several small necrotic pieces were removed for biopsy. Post-operatively the patient developed hypotension. Cardiac tamponade was suspected but a pericardial tap failed to reveal any excess of fluid. The patient died on 6th December at 6.30 p.m. of hepatorenal failure. Bilirubin estimation on 5th December showed 5 mg% with 3 mg of conjugated fraction. Alphafoetoprotein was negative.

Autopsy Reports Post-mortem No. 5518

The wound in the midline of the chest was infected and grew mainly *Staphylococcus pyogenes*.

Liver

Weight, 2,840 gm. There was a large, yellowish-white, spongy area with cavitation in the centre containing putty-like disintegrating material. This occupied the left lobe and part of the right lobe (Fig. 1) compressing the inferior vena cava into a kinked, narrow, tortuous channel (Fig. 2). The base of the right atrium with the inferior vena cava was involved in the necrotic infected mass, and there was no free communication between the right atrium and the inferior vena cava.

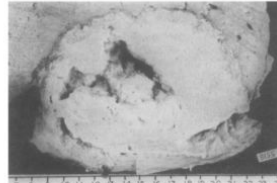


Fig. 1. Cut slice of liver showing large putty-like necrotic mass occupying the left lobe and part of right lobe of liver. There is breaking down in the centre. Bottom shows thickened, adherent friable diaphragm.

A section taken through the mammilated endocardial surface of the right atrium together with the blocked inferior vena cava revealed a necrotic mass of tissue with colonies of Gram-positive cocci and dead alveolar cysts with the classical structureless walls of *E. multilocularis*. The characteristic cysts were numerous in the liver (Figs. 3, 4) and were best seen at the margins of the spongy lesion with the necrotic centre. They were seen to have infiltrated into the surrounding liver tissue. Large radicles of the hepatic vein were thrombosed. The rest of the liver showed areas of passive venous congestion and reversed lobular cirrhosis with complete distortion of the architecture. The portal venous radicles were not involved.

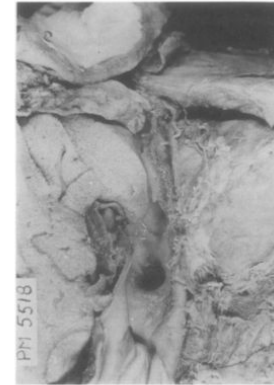


Fig. 2. Intrahepatic portion of inferior vena cava (opened up) compressed by the necrotic mass and becoming tortuous, kinked and almost completely obstructed. Note the thick friable diaphragm above.

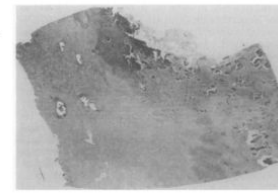


Fig. 3. Photograph of a direct mount of a liver section, showing necrosis on right top with numerous dark alveolar cysts of various sizes and shapes spreading into adjacent liver tissue (PAS).

The base of the left lung, diaphragm and the necrotic mass in liver all seemed to be fused together. Careful slicing revealed that the mass in

the liver had involved the diaphragm and the left basal pleural cavity, giving rise to empyema (100 ml) and also involving the base of the left lung. *E. multilocularis* was diagnosed with inflammatory reaction, considerable fibrosis and passive venous congestion.



Fig. 4. Photomicrograph of necrotic area of liver showing the characteristic dead, irregular-sized and shaped alveolar cysts of *Echinococcus multilocularis*.

Spleen

Weight, 700 g. Histologically it showed the classical features of fibro-congestive splenomegaly.

Heart

Weight 410 g. A purulent exudate was present in the pericardial cavity with numerous Gram-positive colonies of cocci, which, on culture, proved to be *Staphylococcus*. The heart revealed well-established rheumatic heart disease with mitral stenosis and incompetence, tricuspid incompetence and aortic stenosis. Infective endocarditis was observed on the mitral and aortic valves.

Multiorgan Echinococcosis in a Pediatric Patient

Saswati Behera¹ · Monika Bawa¹ · Ravi P. Kanojia¹

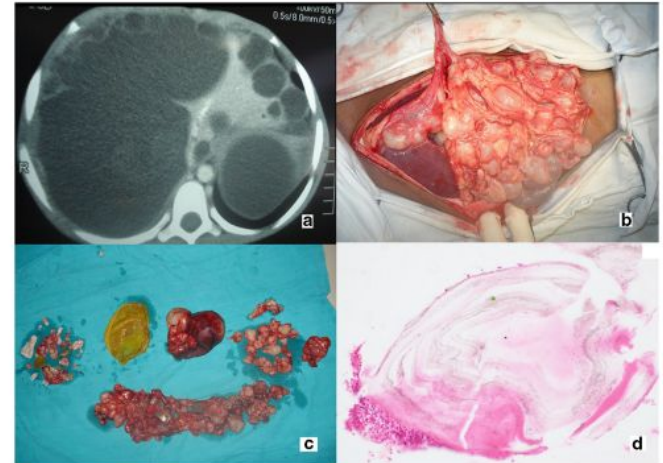
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A ten-year-old male child presented with pain abdomen and distension. Examination showed mild icterus and tender hepatosplenomegaly. Investigations revealed leukocytosis, elevated serum bilirubin, alkaline phosphatase and hydatid serology. Ultrasound and Contrast enhanced computed tomogram showed multiple large cystic lesions in both lobes of liver, spleen and dispersed all over the abdomen (Fig. 1a). There were no cysts in the lungs.

Preoperative oral albendazole (10 mg/kg) was given for 1 month. Intra-operatively, the entire abdominal wall, liver, spleen and omentum were studded with multiple cysts (around 150) (Fig. 1b) which were aspirated and filled with scolicide agent (20% NaCl) for twenty minutes before removing the endocysts to prevent any spillage. Omentectomy and splenectomy were done in view of multiple cysts (Fig. 1c). Histopathological study revealed hydatid cysts with inner germinal, middle laminated and fibrous outer layer with variable inflammation (Fig. 1d). Post-operative bile leak from liver resolved in about four weeks. Albendazole was given for almost an year post-operatively, at intervals, with regular monitoring of liver function tests. Child has no evidence of recurrence at one year follow-up.

Multiorgan involvement of hydatid cysts are rare with only ten cases reported till date in the pediatric age group [1–4]. Two major species, *Echinococcus granulosus* and *E. multilocularis* are of medical importance causing cystic and alveolar echinococcosis respectively. The usual destination of this cyclozoontic parasite is liver via portal vein, however they sometimes reach the right heart and lungs and disseminate to other organs [5]. The symptomatology varies as per the organ system involved. Preoperative imaging reveals unilocular or multilocular cysts with wheel, rosette or honey comb appearance (Fig. 1a) with differentials being amebic and pyogenic liver abscesses [6]. Management continues to be a challenge and aim is to prevent spillage and anaphylaxis intra-operatively. Recurrence due to spillage has been reported in 0–13% [7]. There is no consensus on duration of pre- and postoperative albendazole therapy specially with such multi-organ involvement. Surprisingly, the index patient had no cysts in the lungs which is an unusual scenario as this cyclozoontic parasite involves the liver, lungs and then disseminates to other organs [3].

Fig. 1 a CECT abdomen- Large hydatid cyst completely replacing right lobe of liver with multiple peripheral daughter cysts. Well defined cystic lesions in spleen with dispersed multiple daughter cysts in the entire abdominal cavity. b Omentum can be seen studded with multiple daughter hydatid cysts. c Excised hepatic and omental cysts along with splenectomy specimen. d High magnification depicting the laminated eosinophilic acellular membrane with germinal layer (H&E × 200)



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Mammary Echinococcosis: two Cases and Literature Review

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M. S. Shamim (Departments of Surgery Ziauddin Medical University Hospital* , Karachi.)

Introduction

Hydatid cyst (Echinococcosis) is caused by the organisms *Echinococcus granulosus* and is seen endemically particularly in sheep raising countries. This parasitic infection is common in Middle East and also in other parts of the world including India, Africa, South America, New Zealand, Australia, Turkey and Southern Europe.¹ The disease is manifested by the presence of one or more hydatid cysts usually located in the liver (55-70%) or lung (18-35%) and in both organs simultaneously in 5-13% of the case. Other relatively common sites include muscle (4.7% cases), spleen (2.1% cases) and brain (1.4%). Bone, thyroid, breast and pancreas infections are rarely encountered.¹⁻⁴ In 532 cases reported by Bickers one case each was located in the orbit, bladder, heart, chest wall, sub-cutaneous tissue, tibia, parotid, and thyroid.⁴ Hydatid cyst in the breast usually presents clinically as a painless slowly growing mass without regional lymph node involvement.^{2,5} A retrospective study in the period, 1969-1982 from Salah Aziez Institute, Tunis has reported only 20 cases in the breast.⁶ The breast is a rare primary site of the lesion and accounts for less than 0.27% of all cases.⁷⁻⁹

Case Report

Case 1

A 30 year old female presented with a history of painless mass in the upper outer quadrant of the breast. It ~~was a slow~~ growing lump and had grown to this size in six months. On examination it was a freely mobile soft mass measuring roughly 6.2 x 4.9cm in size. No regional lymph nodes were palpable. The specimen received for histopathology showed a cystic piece of tissue, pearly white in colour and soft in consistency with attached breast tissue flap. Microscopic examination showed a cyst wall composed of lamellated basophilic chitinous material. No scolices were identified, adjacent breast tissue showed extensive secretory change. It was diagnosed as a case of hydatid cyst of breast.

Case 2

The other case was that of a 32 year old female who presented with a small cystic swelling in the breast. It was a painless asymptomatic ~~mass without~~ any axillary lymph node involvement. Clinically there was no suspicion of malignancy. We received a gray white fibrofatty piece of tissue measuring 4x3.5x2 cms in size. On sectioning a cystic cavity was identified measuring 1x1 cm in size. Microscopy showed benign breast tissue along with cyst wall composed of amorphous chitinous material. Several brood capsules containing scolices were also identified. It was also diagnosed as Hydatid cyst. Fine Needle Aspiration Cytology was not performed in both cases.

Discussion

Echinococcosis or hydatid disease, is possibly one of the more difficult parasitic diseases to understand because of the peculiar cystic lesions that form when the larval stages of the parasite invade the viscera. Humans serve as accidental hosts. The normal life cycle of *E. granulosus* involves dogs and foxes as the definitive hosts, within the intestines of which the adult tapeworm reside. Sheep, cattle, or swine serve as the intermediate hosts and develop cystic larval disease.^{10,11} Hydatid cyst can occur in any organ of the body although breast is an uncommon site but a high suspicion of this disease is justified in endemic regions in cases of asymptomatic painless breast lumps without lymph node involvement. Several cases have been reported in the literature, many from developing countries where the disease is endemic. For example cases of mammary echinococcosis have been reported from Turkey^{3,8} and India¹⁰, a case has also been reported from Pakistan.⁹ However, the cases are not confined to developing countries and multiple case reports from developed countries have also been published.^{2,6} The disease can present as a solid or cystic mass in the breast. The solidity is conferred by the fibrous capsule filled with grayish material containing membrane debris. Often a cyst and lump are present together. Primary mammography forms an essential part of pre treatment evaluation since it enables circumscription of the lesion and study of the regional lymph

nodes, which are normal in all cases. Women 20-50 years of age are said to be affected as in our cases both women were in the third decade and of childbearing age. Although rare, several cases are reported to have been diagnosed cytologically by the detection of hooklets in the pleural fluid or sputum. Diagnosing hydatidosis through cytologic examination of aspirated fluid is hazardous, as it can cause anaphylactic reaction due to fluid leak.³ To date 54 cases of hydatid cyst of the breast have been reported and only one was diagnosed through aspiration cytology.⁵ Therefore, although hydatid cyst in breast is a rare lesion, there is enough evidence in literature to justify the need to keep this disease in mind when dealing with mass lesions of breast in the setting of developing countries where the disease is endemic. Surgery remains the main treatment in hydatidosis with well founded criteriae and approach in the most frequent locations of the disease: liver¹¹ and lung.¹² Surgical planning and techniques are dependent on the number of cysts, the anatomical relations and anatomical changes produced by the parasite growth. Surgical approach and technique depends on correct diagnosis and if it has been made before or during operation. The use of antiparasitic medication or solutions during operation varies according to hospitals and medical tradition - hypertonic saline, ether, formic aldehyde, hydrogen peroxide, cetrinide, rivanol and alcohol are commonly used agents.¹³ Chemotherapeutic agents without surgery have demonstrated reduction in the cysts size and occasional elimination of the parasite but not demonstrable benefit has been described so far in large homogeneous series and no scientific conclusions can be drawn.^{14,15} Their mechanism of action is known to be through a blockade of the glucose intake and glycogen deprivation of the parasite with growth retardation and even sterilization of the content. Antiparasitic agents can be used prior to surgery as a safeguard measure and after surgery to prevent further implants and secondary hydatid seeding but not as a sole therapeutic purpose.

CASE REPORT

Open Access



Disseminated alveolar echinococcosis resembling metastatic malignancy: a case report

Laura Caire Nail¹, Ezequiel Rodríguez Reimundes^{1*}, Christelle Weibel Galluzzo², Dan Lebowitz¹, Yasmine Lucile Ibrahim³, Johannes Alexander Lobrinus³ and François Chappuis²

Abstract

Background: Alveolar echinococcosis is a potentially lethal zoonosis caused by larval forms of the tapeworm *Echinococcus multilocularis*.

Humans are aberrant intermediate hosts who become infected by ingestion of egg-contaminated food or water or via physical contact with domestic or wild animals that carry the parasite in their small intestine. In humans, the disease usually affects the liver and can spread to other organs causing metastatic infiltration. In this report, we describe an advanced presentation of human alveolar echinococcosis mimicking metastatic malignancy.

Case presentation: A 62-year-old white woman was evaluated for fever, jaundice, and abdominal pain, associated with significant weight loss. She lived in a rural area in Switzerland and used to eat wild forest fruits and mushrooms. She owned cats that used to hunt rodents.

On physical examination, she appeared severely ill with cachexia, altered mental status, jaundice, and massive hepatomegaly. Laboratory tests showed cholestasis with preserved liver function.

An abdominal computed tomography scan showed an enlarged liver with a huge cystic mass in the right lobe extending into the left lobe, infiltrating her hepatic hilum, causing intrahepatic bile duct dilation and occlusion of her right portal vein. A chest computed tomography scan showed multiple calcified bilateral pulmonary nodules. Her clinical and radiological presentation resembled an advanced neoplastic disease. Serologic tests for *Echinococcus multilocularis* were positive.

The diagnosis of alveolar echinococcosis was established on her past history of exposure, imaging, and serology results.

Conclusions: Clinical presentation and radiologic imaging findings of disseminated alveolar echinococcosis can mimic metastatic malignancy, and diagnosis can be challenging in atypically advanced cases. As the incidence of human alveolar echinococcosis appears to be increasing in Europe and Switzerland, physicians should be aware of alveolar echinococcosis, its epidemiology, and its clinical features.

Keywords: Alveolar echinococcosis, *Echinococcus multilocularis*, Zoonosis, Malignant mimics

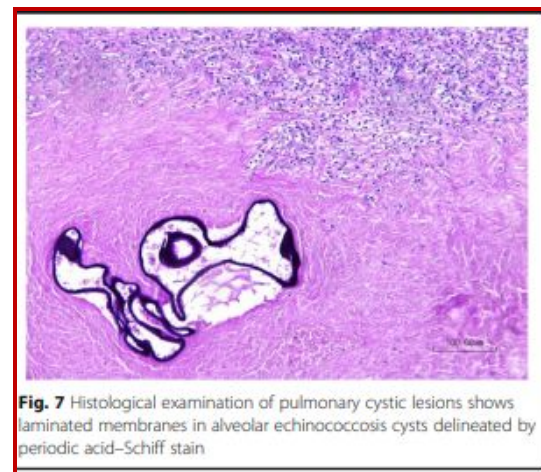


Fig. 7 Histological examination of pulmonary cystic lesions shows laminated membranes in alveolar echinococcosis cysts delineated by periodic acid-Schiff stain

Echinococcosis mimicking liver malignancy: A case report

Radek Pohnan^{a,*}, Miroslav Ryska^a, Vladislav Hytych^b, Radek Matej^c, Petr Hrabal^d, Jiri Pudil^a^a Department of Surgery, Central Military Hospital – Faculty Military Hospital, 2nd Faculty of Medicine, Charles University, Prague, Czech Republic^b Department of Thoracic Surgery, Thomayer's Hospital, Prague, Czech Republic^c Department of Pathology and Molecular Medicine, Thomayer's Hospital, Prague, Czech Republic^d Department of Pathology, Central Military Hospital – Faculty Military Hospital, Prague, Czech Republic

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ABSTRACT

INTRODUCTION: Human Alveolar Echinococcosis – Alveolar Hydatid disease (AE) is an omitted zoonotic infection presenting with focal liver lesions. Cause of AE is a larval stage of *Echinococcus multilocularis* tapeworms.

CASE PRESENTATION: In this report an extraordinary case of a 38-year-old female examined due to 2 liver tumors and 2 pulmonary nodules is described. The patient underwent pulmonary and liver surgery for suspected advanced cholangiocellular carcinoma and surprisingly AE was found.

DISCUSSION: Distinguishing intrahepatic AE from other focal liver lesion can be complicated and in many cases is diagnosed incorrectly as intrahepatic cholangiocarcinoma or other liver malignancy.

CONCLUSION: AE is a rare but potentially fatal parasitic infection primarily affecting liver, although it can metastasise to lung, brain and other organs. The diagnosis and treatment can be difficult and clinical misinterpretation as malignancy is not rare. The principal treatment of AE is surgery accompanied with chemotherapy.

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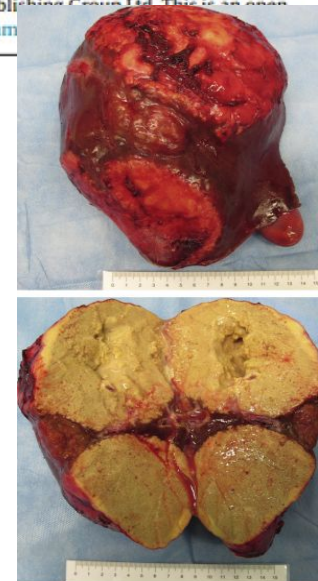


Fig. 3. a,b: Resected right lobe of the liver with tumors.

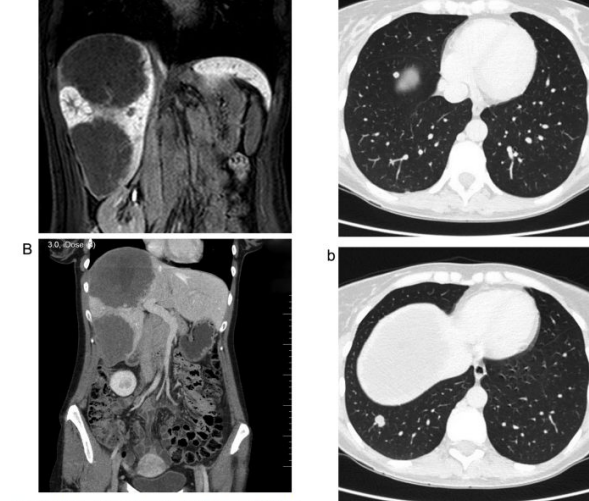


Fig. 1. (a) MRI scan – 2 tumors in the right liver lobe and a mass of focal nodular hyperplasia between the tumors. (b) CT scan – 2 atypical voluminous tumors in the right liver lobe.

Fig. 2. a,b: CT scan – 2 lung nodules in the right lower lobe.

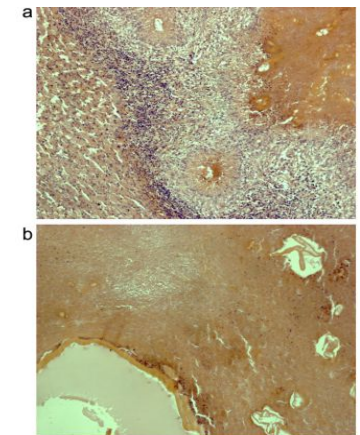


Fig. 4. (a) Extensive necrosis of liver tissue lined with nonspecific granulation tissue (Hematoxyline-eosin, 160x). (b) Necrosis of liver tissue with multiple optically empty spaces (Hematoxyline-eosin, 160x).

and also benign liver lesions such as hemangioma, focal nodular hyperplasia and hepatocellular adenoma [18,19].

In our case there was misinterpretation of advanced cholangiocellular carcinoma based on imaging of infiltrative liver lesions with pulmonary metastasis and false histopathologic diagnosis from liver biopsy, which was initially described as suspected cholangiocellular carcinoma. Later it was reconsidered as necrotic tissue. In 2015 Stojkovic reported that up to one-third of treatment decisions were based on misinterpretation where liver AE was diagnosed

Cystic echinococcosis in the thigh: a case report

Katharina Kurz¹  · Anton Schwabegger² · Stephan Schreieck³ · Bettina Zelger⁴ · Guenter Weiss¹ · Rosa Bellmann-Weiler¹

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Abstract

Intramuscular cystic echinococcosis is a very rare occurrence. Herein we report a case of a 37-year-old patient who presented with progressive swelling of his left thigh. Ultrasound evaluation showed a multicystic, encapsulated lesion (16×3.5×8.5 cm) in the M. vastus lateralis, and serology confirmed the diagnosis of *Echinococcus granulosus* s.l. infection. No additional cysts were detected upon total body CT scan. The patient was treated with albendazole pre-operatively; surgical resection of the mass was then successfully performed. The patient feels well and no signs of residual infestation were seen after 2 years of follow-up.

Keywords Muscular echinococcosis · Thigh · *Echinococcus granulosus* · Case report

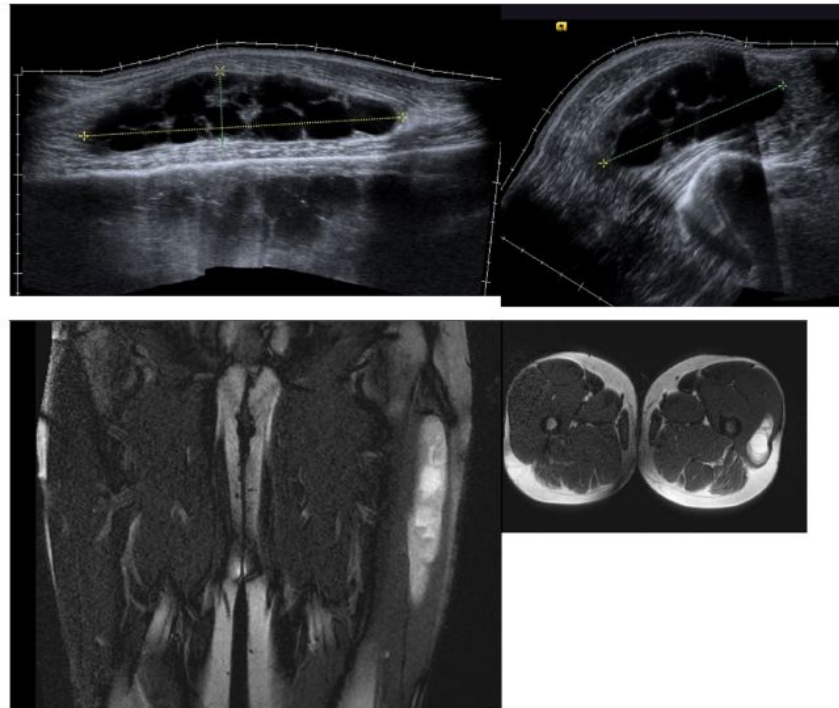


Fig. 1 a + b Ultrasound of the multicystic, encapsulated lesion (16×3.5×8.5 cm) in the M. vastus lateralis of the left thigh. c + d: MRI of the lesion in the thigh (before surgery): albendazole treatment led to discrete regression of the multicystic, encapsulated lesion (16×2.5×8 cm)

Cardiac Alveolar Echinococcosis: a Rare Case Report

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Received: 26 August 2021 / Accepted: 25 September 2021 / Published online: 25 November 2021
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Abstract

There are few case reports in the literature about the cardiac involvement in this tumor-like disease that primarily involves the liver. Although the liver is the most commonly affected organ, other organs may also be affected, including the heart. A 62-year-old male patient, who had been operated three years ago due to hepatic alveolar cyst hydatid, was admitted to the General Surgery clinic with complaints of dyspnea and abdominal pain, and as recurrence was observed in the computed tomography of the patient who had cardiac involvement. In cases with liver AE, other organs must also be evaluated for involvement.



Fig. 1 The appearance of the cyst on Ct

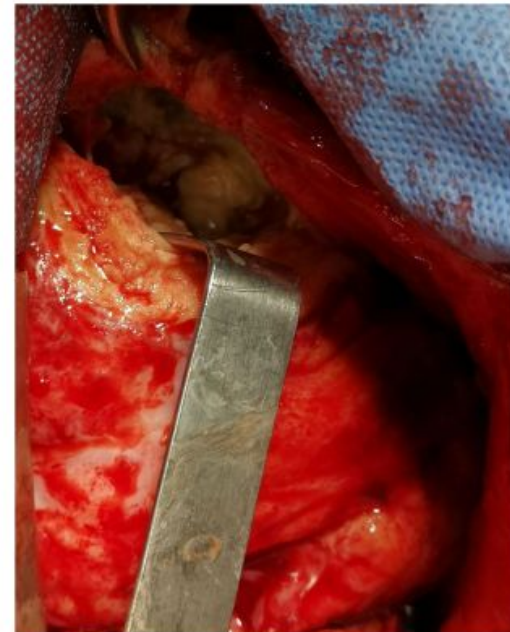


Fig. 4 View during operation

Cerebral alveolar echinococcosis

A case report with MRI and review of the literature

N. ISIK¹, G. SILAV¹, A. ÇERÇİ¹, P. KARABAGLI², I. ELMACI¹, M. KALELIOĞLU³

Alveolar echinococcosis is an important zoonotic infection caused by the larval stage of the *Echinococcus multilocularis*. It is endemic to North America, Central Europe, Russia, China and Turkey. The liver and the lung are the organs most commonly involved. Cerebral alveolar echinococcosis is rare accounting for only 1% of cases. We present a 55-year-old patient with a right frontal mass. T2-weighted MRI series revealed a grape like multilobular, heterogeneous mass with low density. A diagnosis of glial tumor was made. The mass was totally removed. The histopathological examination showed a diffuse growth composed of compartments that are filled with a gelatinous matrix and many brood capsules and protoscolices filled with necrotic tissue. Histopathological findings were consistent with the diagnosis of alveolar echinococcosis. No postoperative complications were observed. There were no lesions in the liver and lungs. The patient was started on albendazole (ABZ) at a daily dosage of 800 mg for 3 months. The patient has remained free of any mass lesion for 5 years. Hypointense grape-like mass with calcification and surrounding white matter edema in T2-weighted MRI should suggest cerebral alveolar echinococcosis. Radical surgery and an adjuvant therapy with ABZ provides useful prolongation of life

Key words: Echinococcosis, surgery - Brain diseases - Magnetic resonance imaging.

Alveolar echinococcosis (AE) or alveolar hydatid disease is an important zoonotic infection caused

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by the larval stage of *Echinococcus multilocularis* that was demonstrated by Rausch and Schiller in 1956.¹ The first alveolar hydatid lesion in the brain was reported in 1884 as being the larval stage of a cestode of genus *Echinococcus*.^{2,3} AE has been reported from Central Europe (France), South America, North America (Canada, Alaska), Australia, New Zealand, China, Russia and Turkey.¹⁻²⁸ AE is almost found in the liver, but may metastasize *via* blood circulation to lungs or other organs. Cerebral AE is very rare accounting for only 1% of cases.^{2,4,6,17,29} According to Bensaid² Clement *et al.*³⁰ reported 37 cases (6%) of cerebral AE among 600 cases collected in Germany and Russia between 1936-1960.² In 1995, Wilson reported 42 active AE cases between 1951-1993 in Alaskan eskimos. Cerebral metastases occurred in 5 patients of them (12%).⁷ In 2000, Bresson-Hadni reported 117 patients with AE.²⁹ The purpose of this report is to present the radiological findings of a right frontal AE with a 5 year surgical follow-up results and review of the surgical cases in the literature.



A rare case of the simultaneous location of *Echinococcus multilocularis* in the liver and the head of the pancreas: case report analysis and review of literature

Marek Kowalczyk^{1,2}, Waldemar Kurpiewski², Ewa Zieliński^{3*} , Dariusz Zadrozny², Łukasz Klepacki², Wit Juśkiewicz², Jacek Lasocki², Łukasz Dyśko², Krzysztof Batia² and Wiesław Pesta²

BMC Infectious Diseases **19**, Article number: 661 (2019) |

Abstract

Background: Echinococcosis multilocularis (Hydatid Disease - HD) is a zoonotic disease caused by the larval form of *Echinococcus multilocularis* (EM). The main sites for this zoonosis are the Middle East, China, India, Alaska, and Siberia. It is rather rare in Europe. In Poland, the Warmian-Masurian Province is the endemic region for *Echinococcus multilocularis*. The clinical manifestation of the disease is dependent on the location, the size of the cyst and the development stage of the parasite. Considering the uncommon character of echinococcosis in Central Europe, especially such located in the areas outside the liver and lungs, the authors would like to present a case of coexistence in one patient of two EM foci in the liver and the head of the pancreas.

Case presentation: We present a clinical case of a 32-year-old man who was diagnosed with a cystic lesion with septa and calcification in the sixth segment of the liver and a suspicious change in the head of the pancreas. ELISA Em 2 plus test was positive, Western Blot method - the P-5 pattern showed an image that is characteristic of an EM infection. The sixth liver segment with a tumour and a tumour from the head of pancreas were excised by means of laparotomy. On the 6th day after the surgery the patient was discharged from hospital without complications and in good condition. Currently, he is under the control of a parasitic and zoonotic clinic. He takes an 800 mg daily dosage of Albendazole.

Conclusions: The presented clinical case shows that if we have a patient with cystic / tumour change in the pancreas and positive immunological tests, CT and MRI of the abdominal cavity are usually sufficient in order to fully diagnose and to qualify such a person for surgery. The most effective treatment is surgical treatment supplemented with pre- and postsurgical treatment with Albendazole.

Keywords: *Echinococcus multilocularis*, Hydatid disease, Pancreas

Echinococcosis in left ventricle: a case report

Li Su, MD, Jianqun Yu, MD, Chengzhong Dai, MD, Yin Liu, MD, Liqing Peng, MD, PhD*

Abstract

Introduction: Echinococcosis, also called hydatid disease, is a common parasitic infection of the liver. However, echinococcus lesions rarely involve the heart, especially in children.

Patient concerns: An 8-year-old child from grazing areas of northwest China was referred to our hospital for the complaint of inpersistent precordial chest pain and left upper quadrant pain for 3 years. Palpation showed hepatomegaly, abdominal palpable mass while inspection abdominal distension. Routine blood tests were within the normal ranges.

Diagnosis: Combining the life history in pasture area, imaging features and serology results, it was consistent with the diagnosis of cardiac echinococcosis.

Interventions: Surgery was performed to evacuate cyst liquid and remove the internal capsule of the cyst.

Outcomes: There was no cystic lesion in heart on ultrasound and her physical condition improved significantly after the surgery. The patient died of hepatic hydatid cyst rupture due to refusing high-risk surgical treatment and other treatment.

Lessons: We presented a rare case of cystic echinococcosis involving left ventricle in a child, and surgery is an alternative and effective therapy for this lesion due to the cyst rupture or leakage that can result in anaphylaxis. The typical imaging features of the cardiac echinococcosis on cardiac magnetic resonance are presented. Patient prognosis relies on proper treatment of all lesions.

Abbreviations: AE = alveolar echinococcosis, CE = cystic echinococcosis, CMR = cardiac magnetic resonance, CT = computed tomography, MRI = magnetic resonance imaging, US = ultrasound, WHO = World Health Organization.

Keywords: computed tomography, echinococcosis, hydatid disease, magnetic resonance imaging, parasitic infection

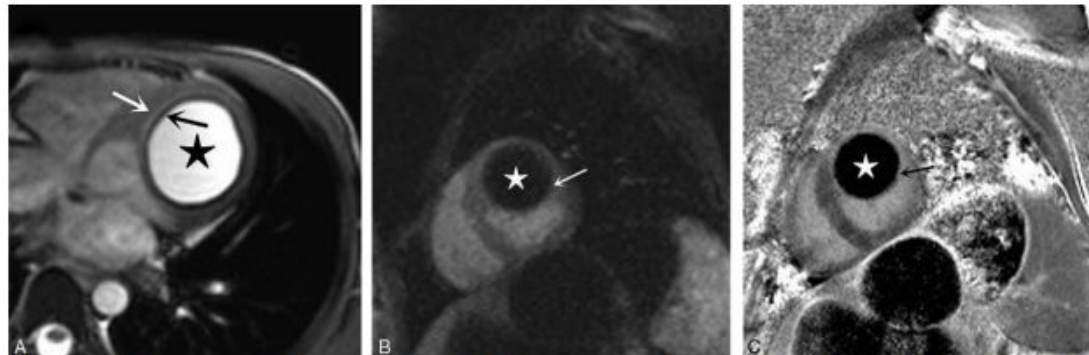


Figure 1. Cardiac magnetic resonance imaging shows the imaging features of the left ventricular echinococcosis (asterisk). (A) A cystic lesion with liquid content and visible cyst wall with "double line sign" (black and white arrows); (B, C) Two chamber view of left ventricle depicts the cyst wall with early contrast enhancement and minimal late gadolinium enhancement (arrow).



Twenty-six years of involvement with cystic echinococcosis: a case report

Hosein Safari¹, Somayeh Mirzavand², Abdollah Rafiei^{2,3} and Molouk Beirumvand^{2,3*} 

Journal of Medical Case Reports **15**, Article number: 266 (2021) |

Abstract

Introduction: Spinal hydatidosis, a zoonotic disease caused by infection with *Echinococcus* spp. larvae, is rare, but its treatment remains a significant medical challenge. Approximately 70% of patients with spinal hydatidosis have lesions in their liver, 0–15% have lung involvement, and only 0.5–2% have bone involvement.

Case presentation: Here we report a 38-year-old Iranian man with spinal hydatidosis, who had a history of eight times surgery in over of 26 years due to hydatid cyst in the liver, lungs, and chest wall. At the most recent admission to hospital he presented with chest pain, paraplegia, and urinary incontinence. Magnetic resonance imaging revealed thoracic spinal hydatid disease. He underwent surgery, and the hydatid cysts were completely removed. Lower extremity forces recovered dramatically and completely within 4 weeks.

Conclusion: Spinal hydatidosis is a rare disease, but it is associated with a high degree of morbidity, mortality, and poor prognosis. Because of the infiltrative nature of hydatid disease, surgery alone is rarely curative. The current case study demonstrates the importance of a suitable surgical approach, adequate intraoperative prophylaxis to prevent cyst rupture, and prolonged complete paraplegia.

Keywords: *Echinococcus granulosus*, Spinal, Cystic echinococcosis

Echinococcosis: an Occupational Disease

M Farahmand,¹ M Yadollahi² www.theijoem.com Vol 1 Number 2; April, 2010

Abstract

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Hydatidosis is a common infestation caused by *Echinococcus* spp. Solitary hydatid cyst of the lung is not uncommon but coexistence of two or more pulmonary cysts are less common. These cysts may drain into the bronchial tree or very rarely into the pleural cavity which causes a poor outcome. Certain people such as slaughters, tanners, stockbreeders, shepherds, butchers, veterinarians and all whose job makes them to work closely with animals are at higher risk of the infection and developing echinococcosis. Herein, we present a 14-year-old shepherd who developed severe chest pain and hydropneumothorax following a minor trauma to his chest. He had two pulmonary hydatid cysts, one of which drained to the left pleural cavity and caused the symptoms. Another cyst was complicated during his hospital course. The patient was treated surgically, received albendazole and discharged home uneventfully. A high index of suspicion is of utmost importance for the correct diagnosis and treatment of hydatid disease in hyperendemic areas and in those whose occupation might put them at a higher risk of contraction of hydatid disease.

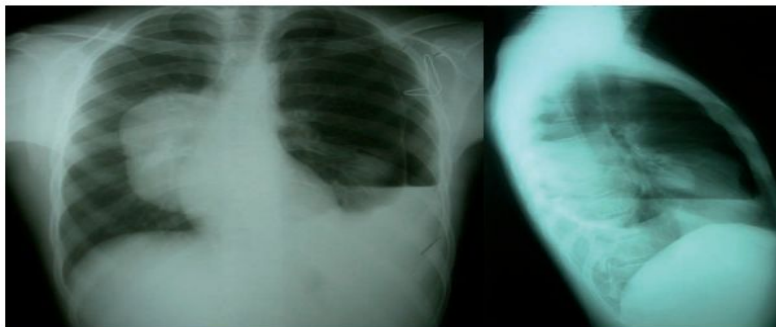


Figure 1: Two large soft tissue densities in both lung fields and hydro-pneumothorax in the left side

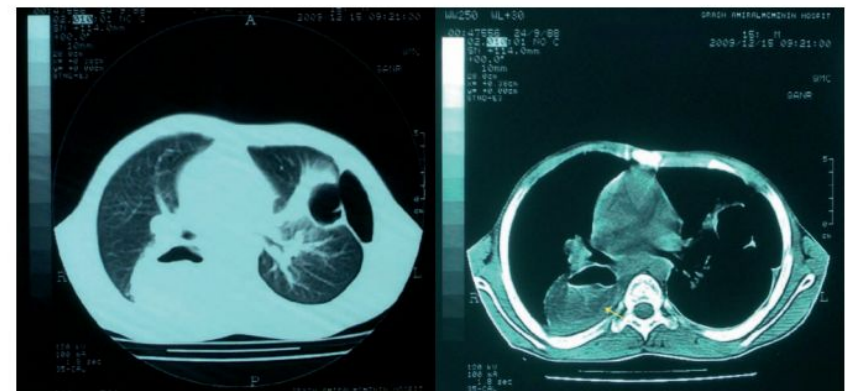


Figure 2: Non-enhanced axial CT showing detachment of the cyst membrane (arrow) in the right cyst producing floating membrane sign. The left cyst caused hydro-pneumothorax and peripheral infiltration.

Case Report: Rare Presentation of Multivisceral Echinococcosis

Xiao Cai,^{1†} Huixia Cai,^{2*†} Qing Gan,¹ Wenxu Chang,¹ Fang Yuan,¹ Wei Luo,¹ Jie Sun,¹ and Jing An¹

¹Department of Otolaryngology Head and Neck Surgery, People's Hospital of Qinghai Province, Xining, China; ²Department of Parasite Control, Qinghai Province Institute for Endemic Diseases Prevention and Control, Xining, China

Abstract. Cystic echinococcosis (CE) is a common, chronic, and endemic zoonotic disease usually localized in a single organ; multivisceral cases are rare, especially outside the liver or lung. Here, we describe an unusual case of a 43-year-old Tibetan man with echinococcosis of the infratemporal fossa, heart, liver, pancreas, abdomen, and pelvic cavity. He only presented with diminished vision of the left eye, especially when chewing. Computed tomography and magnetic resonance imaging revealed multivisceral CE. The patient underwent surgery for the excision of a cyst in the infratemporal fossa, as well as chemotherapy, and the diagnosis was confirmed by histopathological examination. The diagnosis, clinical features, treatment, and follow-up in this case are discussed. In areas with high echinococcosis prevalence, examination by full imaging is necessary for an accurate diagnosis, especially in cases of atypical localization. Chemotherapy for treatment, as well as prophylaxis against recurrence, can be effective when surgery is not possible.

RARE CASE OF MULTIVISCERAL ECHINOCOCCOSIS

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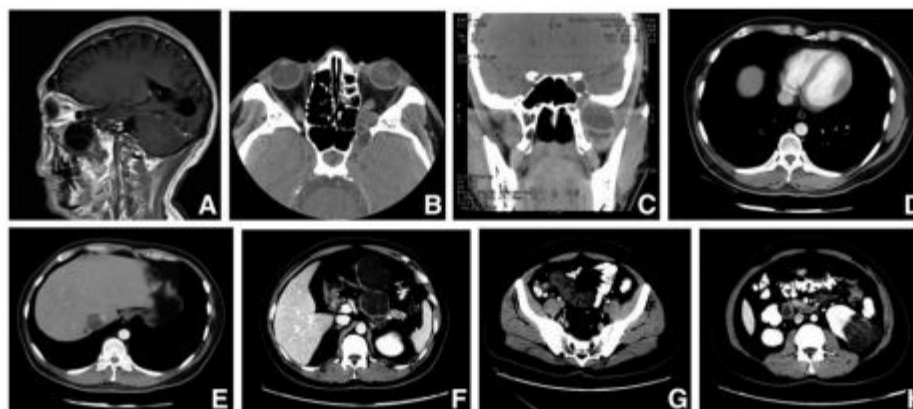


FIGURE 1. (A) Sagittal T2 magnetic resonance image showing the focal lesion with a complete edge and clear separation in the left infratemporal fossa. (B and C) Computed tomography (CT) images showing the cystic lesion infiltrating into the intracranial and orbital areas (CT values: 4 to 50 HU and 3 to 28 HU). (D) Contrast-enhanced CT image showing a low-density mass within the left ventricle (CT value: 12 to 27 HU). (E) Contrast-enhanced CT images showing a cystic lesion within the liver (CT value: -19 HU). (F) Contrast-enhanced CT image showing low-attenuation abdominal cystic masses (CT value: -6 to 18 HU) and a multiseptated mass within the pancreatic tail (CT value: -6 to 21 HU). (G) CT image showing an *Echinococcus granulosus* cyst containing daughter cysts in the pelvic cavity (CT value: -50 to 30 HU). (H) Contrast-enhanced CT image showing a multiseptated thick-walled cystic lesion in the left kidney (CT value: -24 to 27 HU).

Surveillance

- Robust surveillance data is fundamental in order to show burden of disease and to evaluate progress and success of control programmes
- Data is especially scarce and will need more attention if control programmes are to be implemented and measured
- Surveillance for cystic echinococcosis in animals is difficult because the infection is asymptomatic in livestock and dogs
- Surveillance is also not recognized or prioritized by communities or local veterinary services

Human vaccines

- Currently there are no human vaccines against any form of Echinococcosis
- However, there are studies being conducted that are looking at possible vaccine candidates for an effective human vaccine against Echinococcosis.

Vaccines

For Intermediate Host

- *E. granulosus* recombinant antigen (EG95):
 - Remarkable protective efficacy in field trials for sheep
 - The vaccine is currently being produced commercially and is registered in China and Argentina
 - Currently used in endemic areas of China and Argentina
- Gene products of *Echinococcus* oncosphere

For Definitive Host:

- Not currently available
- Gene products of protoscolex or adult worm: suitable candidate
- egM gene family (egM4, egM9, and egM123) in mature adult worms: encouraging protective efficacy in vaccine trials

FUTURE PERSPECTIVES

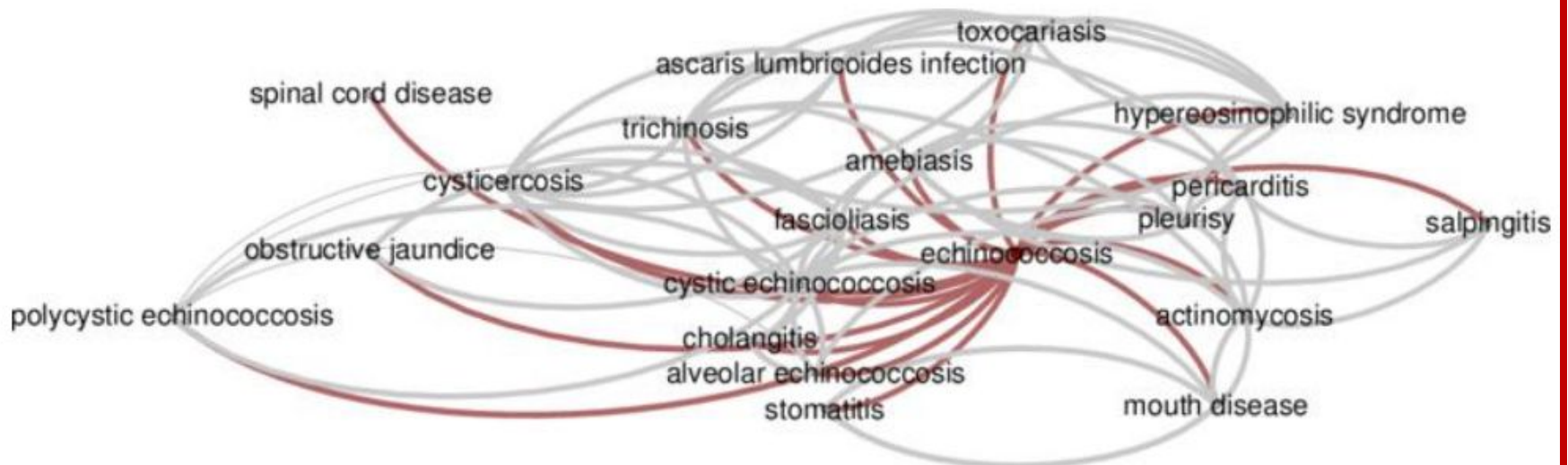
- Under the umbrella of One Health, **WHO** and its partner, the World Organization for Animal Health (OIE) are supporting the development of control programs
- WHO assists countries to develop and implement pilot projects leading to the validation of effective control strategies
- Working with the veterinary and food safety authorities as well as with other sectors is essential to attain the long-term outcomes of reducing the burden of disease

- The first successful control program was initiated in **Iceland nearly 130 years** ago, when hydatid disease was affecting approx one in every six Icelanders (Schantz et al., 1995)
- An extensive health education campaign sensitized the entire population to the disease
- Measures included banning home slaughter of sheep resulting in the gradual elimination of transmission
- By the 1950s echinococcosis was eradicated from Iceland
- Programs initiated in New Zealand (1959) and in Tasmania (1965) were primarily based on education of rural populations and motivating them to change their practices
- Strict control and prohibition of farm slaughter were key features in those programs

- WHO is supporting individual countries to develop control program such as in Mongolia
- In 2018, a multidisciplinary stakeholder meeting was convened in Ulaanbaatar to start developing the **National Action Plan** for control of Hydatidosis
- No significant investment for Hydatidosis has been made, and therefore programmatic steps have been progressing slowly but WHO continues to bring the stakeholders together and further actions have been agreed in 2019
- WHO has also facilitated the validation of diagnostic tests used for Hydatidosis in dogs that is important for surveillance

- One challenge in the control of CE in coming years will be to define optimum targets for developing vaccines effective in definitive canine hosts to interrupt the chain of transmission to humans if Echinococcosis elimination by **2050** by WHO, is to be achieved

Graphical network of the top 20 diseases related to Echinococcosis:



	Cystic	Alveolar	Polycystic	
Agent				
Species	<i>Echinococcus granulosus</i>	<i>Echinococcus multilocularis</i>	<i>Echinococcus vogeli</i>	<i>Echinococcus oligarthrus</i>
Adult size	2-7 mm	4-11 mm	1.9-3.7 mm	1.9-2.9 mm
Definitive host	Dog, wolf, jackal, lion	Foxes	Bush dogs	Wild felid
Intermediate host	Sheep, cattle	Rodents	Paca	Spiny rats
Human disease				
Occurrence	Common	Uncommon	Rare	Extremely rare
Mode of infection	Contact with infected dogs	Contact with fur from infected fox, ingestion of contaminated wild berries	Contact with hunting dogs fed on paca	Contact with infected cats
Lesion appearance	Fluid filled unilocular cyst	Solid mass	Polycystic fluid filled	Polycystic fluid filled
Calcification	Ring type (30%)	Microcalcification or plaque like foci (70%)	Annular, bizarre, amorphous (50%)	Not known
Organs involved				
Primary	Liver (65%), lung (25%), others	Liver (100%)	Liver (100%)	Extrahepatic (orbit others)
Spread	By rupture	Contiguous, metastatic to lung, brain	Contiguous	Contiguous
Presentation	No symptoms, mass effect, rupture, infection	Invasive liver mass, liver failure, cholestasis, Budd chiari, portal hypertension, lung and brain involvement	Liver masses, liver failure, cholestasis, Budd chiari syndrome, portal hypertension	Exophthalmos, soft tissue mass
Mortality untreated (10 years)	Unusual (<5%) (anaphylaxis/complicated cyst)	High (75-100%)	High (>75%)	Not known

Conclusions

- ❖ Neglected tropical disease
- ❖ 2 major species: *E. granulosus*, *E. multilocularis*
- ❖ Human accidental host
- ❖ CE: More common, cosmopolitan
- ❖ AE: less common, more severe
- ❖ Diagnosis: microscopy, imaging, serology, molecular
- ❖ Treatment: surgery, PAIR, chemotherapy, wait and watch
- ❖ Prevention and control: safe animal slaughter, deworming of dogs & vaccination in sheeps

Thank You

