



**Report of the WHO Informal Working Group on cystic  
and alveolar echinococcosis surveillance, prevention and control,  
with the participation of the Food and Agriculture Organization of  
the United Nations and the World Organisation for Animal Health**

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**Department of Control of Neglected Tropical Diseases  
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## **1. Opening session**

Dr Lorenzo Savioli, Director, Department of Control of Neglected Tropical Diseases, welcomed the members of the informal working group on echinococcosis and representatives of the Food and Agriculture Organization of the United Nations (FAO) and the World Organisation for Animal Health (OIE). He extended WHO's thanks to the former coordinator of the informal group, Dr Philip Craig, who had led it for more than 4 years, and to Dr Peter Kern, who had agreed to lead the group for the next 4 years. Dr Savioli thanked all the participants for their dedicated work on a group of parasitic diseases that had a significant public health and economic impact on communities around the world but attracted little attention from decision-makers at national and international levels. He formally opened the Consultation on Cystic and Alveolar Echinococcosis, focusing on the role of veterinary public health in controlling those diseases, the importance of which still is unfortunately often overlooked by both human and veterinary health officers in endemic countries.

Dr François Meslin, Team Leader, Neglected Zoonotic Diseases, said that the purpose of the consultation was to review the current situation of those zoonoses in highly endemic countries, discuss current veterinary public health measures used for their prevention and control, and assess the value of new advances, such as vaccines, and the feasibility of integrated approaches for disease control in intermediate and definitive host species. Dr Meslin also summarized the work of the informal working group on echinococcosis since its creation in 1985 (see section 7).

Dr Philip Craig was nominated Chairman and Dr Thomas Romig Rapporteur of the consultation.

## **2. Updated global burden of cystic and alveolar echinococcosis**

*P.R. Torgerson and P. Craig*

The diseases caused by these parasites represent a substantial burden on the human population. Present estimates suggest that cystic hydatid disease, caused by *Echinococcus granulosus*, results in the loss of 1–3 million disability-adjusted life years per annum. The annual cost of treating cases and economic losses to the livestock industry probably amounts to US\$ 2 billion. Alveolar echinococcosis, caused by *E. multilocularis*, results in the loss of about 650 000 disability-adjusted life years per year. These diseases are perhaps some of the more important global parasitic diseases, with more than 1 million people affected at any one time, many showing severe clinical syndromes.

## **3. Human aspects of cystic and alveolar echinococcosis**

*P. Kern*

The term 'echinococcosis' encompasses two diseases, which differ markedly in their presentation, behaviour and clinical management: alveolar echinococcosis, caused by *E. multilocularis*, in which a multivesiculated tumour forms, mainly in the liver; and cystic echinococcosis, caused by *E. granulosus*, in which fluid-filled cysts (also called hydatids) develop, mainly in the liver or lung. The larval growth of the two parasites separates the

‘malignant’ alveolar echinococcosis from the ‘benign’ cystic form. Experts have recently reached consensus on the diagnosis and treatment of alveolar and cystic echinococcosis, and their recommendations offer guidance to clinicians in areas where these diseases are endemic but rarely diagnosed.

Alveolar echinococcosis is characterized by an asymptomatic incubation period of 5–15 years. Precise diagnosis remains a challenge for clinicians and radiologists, and incomplete knowledge of the disease results in misinterpretation of clinical and imaging findings. Serology is helpful, but, again, clinicians and microbiologists are uncertain about the cross-reactivity of antibodies with antigens from *E. multilocularis* or *E. granulosus*. The PNM classification system<sup>1</sup> is based on imaging findings and has become an international benchmark. Larval metastases may form in organs adjacent to the liver *per continuitatem* or in distant locations by a haematogenous or lymphatic route. To avert the potential ‘malignant’ features of alveolar echinococcosis, continuous larval suppression with benzimidazoles remains the backbone of lifelong treatment. If the lesion is confined, radical surgery offers cure. Unfortunately, in many patients, the disease is diagnosed at an advanced stage, and they undergo palliative surgery without benzimidazoles or with incomplete coverage. As a result, relapses are frequent. Immune deficiency appears to have a strong negative impact according to anecdotal observations. The molecular mechanisms leading to progressive disease are still not known.

Cystic echinococcosis, also known as hydatid disease, is relatively frequent on a global scale. Thus, many clinicians are somewhat familiar with the disease. The incubation period can be as long as that for alveolar echinococcosis, but signs and symptoms are often reported within a shorter period, depending on the location of the cyst(s). Besides the liver and lung, other organs can be affected. In contrast to alveolar echinococcosis, the larva forms a single or several fluid-filled cysts—known since ancient times as hydatids, which are always surrounded by a well-organized, compact capsule of host origin. Larval growth occurs inside the cyst (endogenous budding). Young, unruptured cysts often remain serologically negative. The WHO system for classifying hepatic cysts by ultrasound imaging has also become the benchmark,<sup>2</sup> and its application has much improved both the clinical management and international comparison of clinical data. Experience with interventional procedures, such as percutaneous puncture, is encouraging, and use of this technique for hepatic cysts has been shown to be safe. Surgery may no longer be the treatment of choice; instead, many centres use short-term cycles of benzimidazole, which degenerates the endocyst and results subsequently in a regressive course of cystic echinococcosis. Others favour the ‘watch and wait’ principle and carefully observed natural degeneration of the cyst. Diagnosis of regressive cystic echinococcosis is, however, still a challenge; serology is not well adapted for identifying either the remnants or the viability of the parasite, and imaging findings result in unnecessary surgery, exposing the patients to high risk,

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<sup>1</sup> The WHO PNM classification system covers a wide clinical spectrum, with four ‘P’ categories for the distribution of lesions (P for parasite in the liver), two ‘N’ and two ‘M’ categories for the presence or absence of local infiltration including lymph nodes (N) and metastasis (M), respectively. Stages I–IV are derived from those categories.

<sup>2</sup> Annex 1 in *PAIR: puncture, aspiration, injection, re-aspiration: an option for the treatment of cystic echinococcosis*. Geneva, World Health Organization, 2001 (WHO/CDS/CSR/APH/2001.6).

particularly in regions where both cystic and alveolar echinococcosis are diagnosed. Consultation at specialized centres is recommended.

#### **4. Veterinary public health and veterinary surveillance**

*F.X. Meslin and P. Craig*

Veterinary public health is listed as one of the five strategies for overcoming neglected tropical diseases in the report entitled *Working to overcome the global impact of neglected tropical diseases*<sup>1</sup> launched by the WHO Director-General on 14 October 2010. In her speech, Dr Chan acknowledged the importance of veterinary public health, particularly for the prevention and control of zoonotic diseases. This Neglected Tropical Diseases report, which was well received by the public health and donor communities, provides technical information on 17 diseases, including three zoonoses: porcine cysticercosis (taeniosis), human and dog rabies and cystic echinococcosis (hydatidosis). Inclusion of these infections on the Neglected Tropical Diseases list has raised interest and should help to improve the visibility of both cystic and alveolar echinococcosis.

Annual surveillance of infection rates in dogs, livestock and humans is critical for establishing a pre-intervention baseline, assessing the efficacy of control programmes and providing data to control authorities and organizations for forward planning and budget approval. Purging of owned dogs with arecoline and necropsy of strays may largely be replaced by testing of faeces with a coproantigen enzyme-linked immunosorbent assay. Livestock slaughter inspection records (from local slaughterhouses or purchased animals) remains the gold standard, but cysts <1 cm require histological verification or DNA analysis. Livestock serology is of limited use—probably only for imported or exported live animals and possibly for herd testing for consolidation or maintenance of eradication phases. Surveillance tools are widely available for control of *E. granulosus*.

#### **5. Review of control programmes and disease epidemiology**

##### **5.1 Control of *Echinococcus granulosus* and cystic echinococcosis**

###### **5.1.1 General aspects**

*P. Craig*

The disease caused by *E. granulosus*, cystic echinococcosis, is one of the neglected zoonotic diseases recognized by WHO. It represents a significant global human disease burden in resource-poor pastoral communities. Transmission occurs from dogs to humans but not directly between humans. Interventions should therefore be targeted to animal hosts; however, both definitive (dogs) and intermediate animal hosts (sheep, other livestock) are asymptomatic, and cystic echinococcosis is not of significant economic importance to livestock owners or governments. The chronicity of human cystic echinococcosis, fragmented medical records, its

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<sup>1</sup> *Working to overcome the global impact of neglected tropical diseases*. Geneva, World Health Organization, 2010 (WHO/HTM/NTD/2010.1).

complicated diagnosis and difficult and costly treatment result in further neglect. Interventions to reduce the zoonotic risk and human cases are necessarily directed to treating domestic dogs and to changes in husbandry, which require veterinary action and surveillance. Control should be a concern of public health rather than animal health; however, it has low priority and intersectoral cooperation is often poor. An active programme for cystic echinococcosis case management, treatment and follow-up is vital. Hydatid control programmes begun during the 1960s and 1980s indicate that effective intervention is possible with established tools but requires time (more than 5–10 years).

Supervised treatment of owned dogs four to eight times a year with praziquantel is the single most important intervention. Its effectiveness depends on the percentage of the dog population registered and treated, access to households, the efficiency of the dosing team, community acceptance, logistics and management. Other interventions include changing livestock husbandry and home-slaughter practices, meat inspection and dog management. Health education can increase participation and maintain the continuity of long-term control, especially in the consolidation phase. Vaccination of sheep with EG95 vaccine should reduce transmission to dogs and the time required to achieve control. Control end-points should be defined. Simulations of animal prevalence and frequency distributions, intervention parameters, cost–benefit and other factors should be modelled before and during control. Veterinary surveillance is described in section 4; medical surveillance, from hospital records (surgical, medical, outpatient), provides a basis for health impact assessment, with community-based mass ultrasound examinations annually, especially among people aged <15 years. Human serological screening alone is not always helpful and may result in large numbers of false-positives for long-term follow-up. Recognized hydatid treatment centres should be identified in endemic areas, in which surgical, chemotherapeutic, PAIR (puncture, aspiration, injection, re-aspiration) and ‘watch-and-wait’ approaches are available (for in- or outpatients) for the various clinico-pathological presentations.

The success of a hydatid programme often depends on the structure, effectiveness and sustainable funding of the control authority and team and the willingness of target communities to participate, rather than on the technical tools and approaches available. Integrated dosing of dogs with praziquantel (at a defined frequency) combined with vaccination of livestock with EG95, with effective surveillance, have not yet been adequately assessed. Guidelines for successful control require clarification in relation to attack, consolidation and maintenance of elimination phases, and best practice must be identified for monitoring the parasite in transboundary situations. Identification of exposure of young livestock and children to cystic echinococcosis during and after control is difficult. An effective vaccine against *E. granulosus*

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