

ARTIGOS

POLYCYSTIC HYDATID DISEASE IN BRAZIL. REPORT OF FIVE NEW HUMAN CASES AND A SHORT REVIEW OF OTHER PUBLISHED OBSERVATIONS

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This paper describes five additional Brazilian human cases of polycystic hydatid disease due to Echinococcus vogeli, reviews the previous cases reported in Brazil, including one report of E. oligarthus (20 in total), and some epidemiological aspects of this disease which is no longer a curiosity but rather a problem that is not medically easy to handle. Its presence should be expected in any rural area of the New World where humans have not eliminated wild felids/canids, bush dogs, pacas, agoutis and other wild rodents.

Key-words: Polycystic hydatid disease. Echinococcus vogeli. Brazil.

Echinococcus granulosus is known to be endemic in humans, ungulates and domestic dogs in Uruguay, Argentina, Chile, the Andean zone of Peru and Rio Grande do Sul, Brazil. However, the parasite probably is present in every country in the continental Americas⁵, but sporadic human cases are rare or seen in immigrants from endemic areas of the same or another country. The reason for this is not clear, but it may be due to epidemiological circumstances that prevent transmission, or to the behavior of the strain of the parasite present in non-endemic areas.

In addition, hydatids showing a polycystic morphology, rather than unilocular, had been observed in humans and animals and were cataloged as alveolar, multicystic or multilocular hydatids and interpreted as due to *E. granulosus*, *E. multilocularis* (cycle canid-small rodent, in the arctic zones) or more recently to *E. oligarthus* (wild cat-agouti cycle, mostly in tropical areas). However, these diagnoses were not substantiated by experimental infections in carnivores to obtain the adult worms which are easy to differentiate.

Fortunately, while working in Colombia, we were able to infect dogs and cats with polycystic hydatid metacestodes of human and animal origin and demonstrated the presence in the country of the two neotropical *Echinococcus* species *E. oligarthus* and *E. vogeli* (Ev). We described the larval characteristics of Ev, also polycystic, in the only important known host, the paca (*Cuniculus paca*). The adult parasite had been described by Rausch and Bernstein²⁶ in a bush dog (*Speothos venaticus*) from Ecuador and, as they had predicted due to the food habits of the bush dog, the intermediate host was found to be the paca.

As mentioned above, experimental infections demonstrated that the metacestodes of the two neotropical species were both polycystic. The differential morphological characteristics were studied and described. It was found that the shape, the relative proportion of handle and guard/blade and the size of the hooklets of the protoscolices were of particular importance^{7 8 28 29}.

Since the original description of polycystic hydatid disease (PHD) in 1979 involving 13 cases in 4 countries, no less than 63 human cases due to Ev (and also *E. oligarthus*) have been found in 10 countries, including Brazil.

This paper describes the information available (mostly pathological) on 5 additional Brazilian human cases of polycystic hydatid disease (PHD) and discusses the other cases reported in Brazil (Table 1), and some epidemiological aspects of this interesting disease which is no longer a curiosity but rather a problem not medically easy to handle.

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Recebido para publicação em 20/06/95.

Table 1 - Brazilian human cases of polycystic hydatid disease.

Number	State	Age	Sex	Clinical and imaging highlights	Evaluation time	Location	Hydatid	Treatment	Follow-up	Ref #
1	Goiás,	31	F	Wgt loss, jaundice enlarged abdomen, portal hypertension, esophageal varices	8 m	liver	Ev	3 bile obstructed duct, surgically drained through colecystectomy	massive hemorrhage,	pp*
2	Pará	45	F	Unknown	?	liver	Ev	biopsy	hematemesis, shock, death	pp
3	Anapó	67	M	Undescribed illness	7 y	liver colon	Ev	surgical excision of PHID	unknown	pp
4	Pará	51	F	Unknown	?	liver, omentum	PHID	4 surgical interventions	unknown	pp
5	Pará	56	M	Diagnosed PHID in 1980	?	liver	Ev	biopsy	died in 1987, no autopsy	pp
6	M. Gerais	70	M	Died of tetanus	?	heart	Ev	none	autopsy finding	6
7	M. Gerais	22	M	Progressive wgt loss, anemia, pain RUQ, jaundice, hepatosplenomegaly, esophageal varices.	20m	liver, diaphragm, omentum	Ev	exploratory surgery, mebendazole for 12 m	died, biliary cirrhosis	11
8	M. Grosso Sul	60	F	US: multiple intrahepatic cysts Wgt loss, anorexia, anemia, previous surgery showed PHID.	13 y	liver, spleen	Ev	none	died, autopsy: PHID	9
9	M. Grosso Sul	44	M	US: multiple intrahepatic cysts 20kg wgt loss/2m, obstruction bile system. CT showed many liver cysts	?	liver	PHID	exploratory surg., albendazole 10mg/kg/d x 3m	marked improved, wgt. regained, smaller cysts at CT	27
10	Pará	43	M	Wgt loss, jaundice, hepatoma-like. Linear liver calcification at x-ray	6 m	liver	PHID	at surgery PHID blocking biliary system	at autopsy most liver replaced by PHID	24
11	Pará	12	F	Intraumbilical mass, irregular hepatosplenomegaly	?	mesentery base	PHID	surgical removal PHID	undetermined	22
12	Acre	20	M	29km wgt loss, progressive enlargement abdomen due to painless growing intraabdominal masses	5 m	liver, mesentery, pancreas, spleen, pelvis	Ev	surgical removal PHID, albendazole x 6 m	at 1y imaging neg., clinically well	18
13	Acre	30	F	Jaundice, enlarging, painful nodules in epigastrium, hepatosplenomegaly, esoph. varices. US, CT showed many cysts and calcification in liver	3 y	liver	Ev	biopsy, albendazole intolerance, 50 days treatment	unknown	20
14	Acre	35	M	Anemia, jaundice, enlarging painful nodules in epigastrium, irregular hepatosplenomegaly, esoph. varices, high eosinophilia. Simple x-ray, US, CT, showed multiple cystic nodules	3 y	liver	PHID	biopsy, albendazole	success, details not available	20
15	Acre	48	F	Wgt loss, abdominal pain, vomiting. US, CT showed no lesion in liver, spleen, pancreas	2 y	mesentery	PHID	surgical removal	apparently cured	20
16	Acre	58	F	Pain in upper chest, epigastrium, dry cough, dyspnea, hemoptysis. X-ray showed multiple cancer-like nodules in both lungs	3 y	lung, liver	PHID	biopsy, albendazole x 3 m	at 1 y residual lesions in lungs, calcified lesions in liver	20
17	Acre	34	M	10kg wgt loss, pain in RUQ, jaundice, hemoptysis. Hard, irregular hepatomegaly, esoph. varices. US, CT showed rounded masses in chest and liver	5 y	liver, lung	Ev	biopsy: albendazole	stop hemoptysis, jaundice continue, 20 surgery to clear bile drain failed, 2 y later, good geral condition, cirrhosis symptoms unchanged	20, 21
18	Acre	38	F	Intermittent jaundice, mass in RUQ, hard nodules, enlarged liver, eosinophilia. US & CT showed cystic lesions and sprinkled calcifications	?	liver	Ev	biopsy, albendazole	"success"	20, 21
19	São Paulo, Bahia,	76	M	Asymptomatic, painless, round calcified masses in the liver	?	liver	Ev?	none	asymptomatic	20
20	São Paulo, Bahia,	54	F	Asymptomatic for 20 y after episode of cholecistitis-like. Painful, hard hepatomegaly. Chest and abdomen x-ray: disseminated small calcified nodules	?	liver, lung	Ev?	none	asymptomatic	17, 20

* = present paper; abbreviations: CT = computed tomography, US = ultra sound, RUQ = right upper quadrant, PHID = polycystic hydatid disease, m = months, y = year

Polycystic hydatid infections in Brazilian animals. Brumpt and Joyeux² found in the liver and spleen of 1 of 4 agoutis (*Dasyprocta agouti* = *D. leporina*) collected in Alburquerque, Lins, State of Sao Paulo, a polycystic hydatid that they described as the metacestode of a new species of *Echinococcus*: *E. cruzi* rather than *E. oligarthrus*. Based on a careful study of the differential characteristics of the 2 neotropical species, Ev and *E. oligarthrus*, from different origins and the original slides of *E. cruzi*, it was arrived to the conclusion that the rostellar hooklets from protoscolices of *E. oligarthrus* and *E. cruzi* corresponded in form, although those from the latter were slightly greater in length. The difference was considered of no taxonomic significance and therefore they placed *E. cruzi* in synonymy with *E. oligarthrus*²⁶.

The occurrence of the larval stage of *Echinococcus* spp in the Americas by species of host and country were summarized⁸. In Brazil, other metacestodes collected in agoutis near Belem were also found to be *E. oligarthrus* but the report of *E. granulosis* in a Brazilian agouti by Lutz lacks sufficient information to allow a definitive taxonomic opinion. In addition to these observations, *E. oligarthrus* has been reported in various countries and in a few species of mammals: *Proechimys* spp. (spiny rat), and once in *Cuniculus paca*, and in *Didelphis marsupialis* (opossum). More recently Ev was found in *Sylvilagus floridanus* (wild rabbit) from Venezuela¹⁶ and in pacas from Para State (Moraes, this report); in Acre State²¹; in Bolivia¹² and in Ecuador¹⁴.

The carnivores reported naturally infected with *E. oligarthrus* are the following wild cats: *Felis yagouaroundi*, *F. concolor*, *F. onca*, *F. pardalis*, *F. geoffroyi* and *F. colocolo*⁸; several of these cats have extensive geographic ranges. The widest is that of the cougar, *Felis concolor*, which occurs in northern British Columbia, Canada, and southward to Tierra del Fuego. Some, such as the ocelot and the jaguar, which formely were present in SW the US seem to have disappeared from this country. On the contrary, the bush dog, *Speothos venaticus* (Figure 1), distributed from Panama to northern Argentina, and the domestic dog, are the only animals that have been reported naturally infected with Ev and

the paca is the only important intermediate hosts (Figure 2).

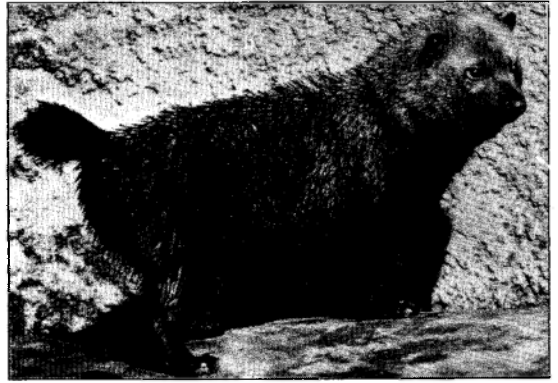


Figure 1 - Bush dog, *Speothos venaticus*. Photo courtesy of the Los Angeles Zoo.



Figure 2 - Adult paca, *Cuniculus paca*. Photo courtesy of Dr. Ignacio Borrero, Universidad del Valle, Cali, Colombia.

It should be mentioned that PHD transmission has been reported in zoos where bush dogs served as a source of infection for nutrias, *Myocastor coypus*⁹, and a group of primates: gorillas, orangutans and chimpanzees¹⁵²³. This information should be useful to alert zoo veterinarians to detect infections in captive bush dogs, using antiparasitic drugs to recover the adult worms and in primates, using ultrasound procedures to detect the metacestodes.

Diagnosis. It seems pertinent in this short review to mention how the diagnosis of PHD is carried out in humans.

1. Demonstration of polycystic masses by physical examination or by using imaging

procedures: x ray, showing polycystic tumors, usually with some calcifications 2-3cm or more in diameter with irregular borders, but they may have diverse forms and configurations. Ultrasound, CT and MRI are more sophisticated and expensive procedures but useful for diagnosis^{19,22}. The metacestode is usually present simultaneously in various organs of the upper and lower abdomen, the chest, and the orbit (D'Alessandro, unpublished).

2. The patient lives or has lived in rural areas with abundant wild life, and usually is familiar with pacas.
3. Serological tests. IHA and Immunoblot usually are positive in cases diagnosed by other means. Very recently, Gottstein et al 1995¹³ have obtained a purified Ev antigen, Ev2, allowing discrimination of non-*Echinococcus infections* and *E. granulosus* infections from Ev. Only some alveolar hydatid cases could not be discriminated. However, the distribution of *E. multilocularis* is holartic rather than neotropical so its distribution does not overlap in these regions.
4. Parasitological characteristics of PHD obtained by biopsy, pathological specimens or necropsy: a) the shape, proportions of the parts of the protoscolex hooklets (better seen in squash preparations) and b) the morphology of the body or walls of the hydatid in tissue sections stained with H & E and PAS²⁹.

Case Reports

Case 1. (MVS (A93-87). A 31-year-old Brazilian female agricultural worker, born in Goias State but living in Mato Grosso at the time of presentation, was admitted to the teaching hospital of Brasilia in October 1987 complaining of an enlarged abdomen, diarrhea, increasing jaundice, and generalized itching since a few months. She had been diagnosed as having viral hepatitis one month earlier in another hospital. At admission she was found to be in poor general health, with a history of a 12kg weight loss during the previous weeks. She was markedly jaundiced, and the physical exam revealed palmar erythema and signs of generalized skin lesions due to scratching. The abdomen was distended and the liver was hard and palpable

20cm below the right costal margin. No ascites or abdominal collateral circulation were detected. Main laboratory findings at admission included: total bilirubin, 33mg/dl with the direct fraction of 21mg/dl; alkaline phosphatase, 57U; AST 36U; ALT 40U; total protein 6.8gr/dl, albumin 3.2gr/dl; creatinine 0.7/dl. The WBC count was normal but 11% eosinophils was reported. The abdomen ultrasound showed enlarged liver and spleen with a cavernous mass in the porta hepatis, dilated intrahepatic bile ducts, signs of portal thrombosis, and a calcified nodule in the liver (2cm x 2cm). With the diagnosis of obstructive jaundice and portal hypertension, an exploratory laparotomy was carried out one month after admission. Examination of the porta hepatis showed enlarged, recanalized umbilical veins. The gallbladder appeared shriveled and without calculi. When the common bile duct was opened, multiple small vesicles were seen (Figure 3). An intraoperative cholangiogram revealed the left hepatic bile duct to be obstructed. The right



Figure 3 - *Echinococcus vogeli* cyst obtained from the liver and the biliary drain of case 1. Note the diversity of size and the presence of brood capsules, free and within the cysts. Bar = 1cm.

hepatic bile duct was impermeable and the cystic duct was fibrotic. A cholecystectomy was performed and external drainage of the biliary system was established. Two days following surgery an episode of bleeding from the surgical excision and the externalized drain was observed, and fluid from the drain contained parasitic cysts. Two weeks after surgery the patient experienced massive hematemesis, hypovolemic shock, and died the following day. At autopsy numerous vesicles were observed on the surface and in sections of the parenchyma and the porta hepatis of the liver (Figure 4). The parasite was alive in some areas and dead in others. The

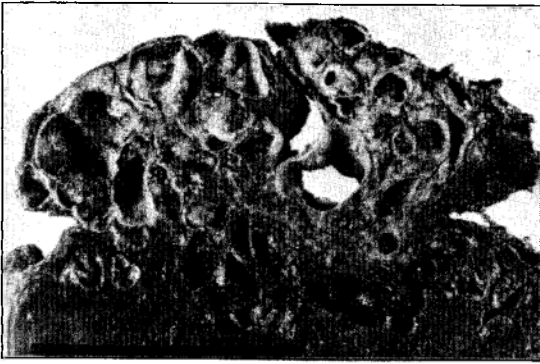


Figure 4 - *Echinococcus vogeli* shown in a liver section of case 1. The polycystic appearance of the lesion can be clearly seen as well as the invasion of the parenchyma by the metacestode. Bar = 20cm.

laminated membrane was thick and the germinal thin, showing few calcareous corpuscles. Brood capsules and protoscolices with typical Ev hooklets were frequently seen. (Figure 5 and 6). Between the host fibrous tissue reaction and the parasite, an accumulation of granular material representing dead eosinophils was frequently seen. In several areas proliferation of the laminated membrane was evident, at times having a cerebriform appearance. In some protoescolices several abnormal looking hooklets were seen (Figure 7).

Case 2. (OAF, Brasilia University, BU-29023) Year 1976. A 45 year-old female from Curralinho, rural area of Para State, with no clinical history available. A liver biopsy contained a polycystic hydatid with a laminated membrane showing a cerebriform appearance (Figure 8). The parasite was

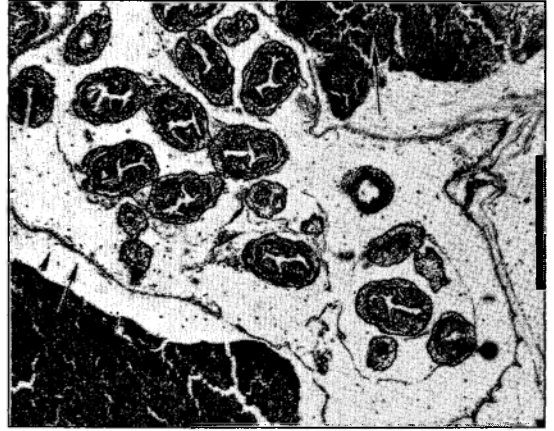


Figure 5 - Tissue section (H & E) of *Echinococcus vogeli* of case 1. Thick laminated membrane (arrowhead), thin germinal membrane (medium length arrow), granular material between the laminated membrane and the liver (long length arrow), brood capsules and protoscolices. Bar = 200µm.

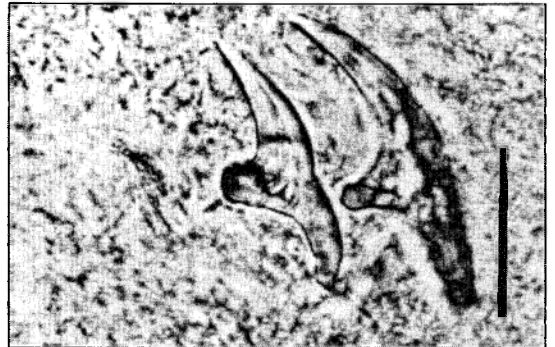


Figure 6 - Unstained squash preparation of fixed protoscolices of case 1, showing large and small hooklets of *E. vogeli*. Bar = 20µm.

mostly dead and surrounded by histiocytes arranged in a palisade fashion. Protoscolices and hooklets conformed to those of *Ev*.

Case 3. (DMB, BU- 28000). Year 1976. A 67 year-old male from Macapa, Amapa State. Tumors of the liver and of colon were excised after 7 years of illness. The metacestode was dead but it showed *Ev* hooklets and proliferating, cerebriform laminated membrane.

Case 4. (MSS, BU-13539). Year 1971. A 51 year-old female from Moju, Para State, underwent surgery 4 times, 3 of which antedated 1971. During the first surgery,

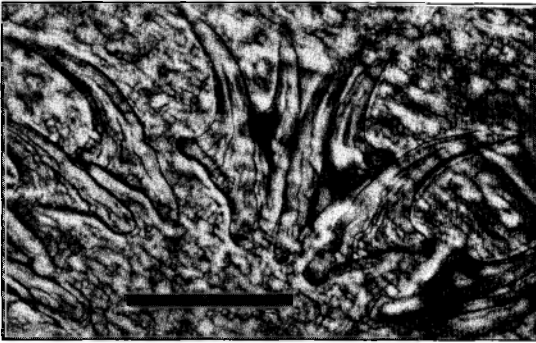


Figure 7 - Unstained squash preparation of fixed protoscolices of case 1, showing normal and abnormal hooklets (arrows) of *E. vogeli*. Bar = 20um.

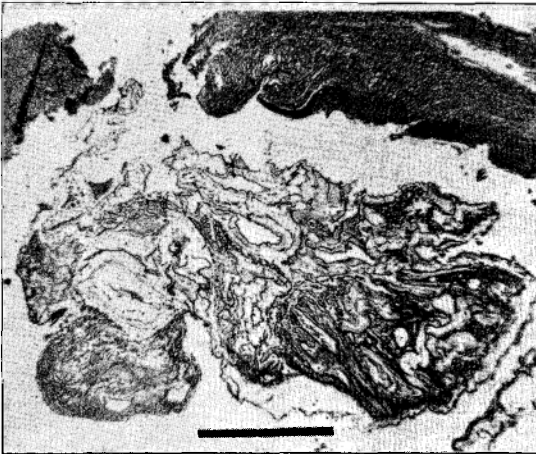


Figure 8 - Tissue section (H & E). Convoluted laminated membrane within the lumen of a cyst (case 3) given the appearance of a cerebroid formation. Bar = 20um.

polycystic lesions from the liver and omentum were removed. This is the only information that was obtained.

Case 5. (BSR, studied by Mario Moraes in 1987). A 56 year-old agricultural worker from Paragominas, Para State. In 1980 he had been told that he was infected with hydatid disease. The patient died in 1987 and an autopsy could not be performed. However, specimens available from previous biopsies showed he was infected with polycystic hydatids with a laminated membrane showing a cerebroid appearance. Protoscolices and Ev hooklets were seen. This patient had brought with him

an Ev-infected paca to demonstrate the parasitic lesions of the animal.

In summary, five new cases of PHD are reported. In 4 the characteristics of the metacestode and, in particular, the protoscolices' hooklets indicate that the infection was due to Ev. The first patient died of complications after surgery carried out to allow the bile to drain. Little clinical information was available from the other 4 cases except that the illness that originated the surgery to make a diagnosis demonstrated that it was a PHD and that the parasite involved in at least 3 cases was Ev due to the morphology of the hooklets. Whether the cerebroid appearance of the laminated membrane of the 4th case is sufficient to indicate that it is due to Ev is a point that should be confirmed by the study of more *E. oligarthrus* in humans.

A summary of the data of the new and previously reported human cases in Brazil is presented in Table 1.

DISCUSSION

As stated earlier, it is understandable that PHD was not diagnosed until recently. Ev was described in 1972²⁶ in the bush dog, but the metacestode was not recognized until the observations made with Colombian material^{7,29}. With the differential criteria established, it became possible to recognize that Eo was the origin of an Echinococcus heart infection found outside the endemic area of *E. granulosus* in Brazil⁶. Also that cases 2 and 3 in the present report, recognized in 1976 as polycystic and therefore not due to *E. granulosus*, were not published by Moraes up until now, in the present report. They were both Ev. Reports of all Brazilian cases, including those presented in congresses (as new, overlooked, or previously unreported), are summarized in Table 1.

So far, in Brazil, there have been 20 PHD cases reported, 9 males and 11 females; the mean age was 41 with a range of 12 to 76 years. Only 7 were younger than 30. Three cases were asymptomatic individuals; 2 with groups of rounded calcifications in the liver and lung (considered probably dead and calcified metacestodes of PHD) and the patient with *E. oligarthrus* in the heart, who died of tetanus and at autopsy showed this parasite as an accidental finding.

The location of the parasites was: liver alone (9 cases), liver and lung (5 cases), base of the mesentery (1 case) and cysts in the liver and other organs in the last 4. Five of these 20 cases died as a consequence of the PHD which had caused liver cirrhosis, obstructive jaundice, portal hypertension and or esophageal varices.

Of the 20 cases, 11 were definitely due to *Ev* and one to *E. oligarthrus* because the typical characteristics of the hooklets; in 7, however, hooklets were not seen and in 2 there were no pathological specimens available for microscopic examination (only radiological). Although the typical characteristics of the *E. oligarthrus* metacestode is well established, only one case of a dead *E. oligarthrus* in a man's heart has been described showing not only hooklets but its cystic appearance, not different from that shown in animals.

It is clear that *Ev* is found much more frequently in humans than is *E. oligarthrus*. The former is transmitted from the domestic dog, which is a good host for *Ev* but not for *E. oligarthrus*; so far as has been observed, the latter develops to sexual maturity and produces eggs only in wild cats (Felidae) of various species⁸.

Fortunately, most of the Brazilian investigators have been aware of the studies of PHD, and have recognized its presence in Brazil. In 1992 it was found an endemic area of human transmission in Acre State²². The patients were thoroughly studied and longitudinal observations made possible the evaluation of beneficial response to albendazole treatment in 4 of 6 patients²⁰. On the other hand, 1 year of mebendazole treatment was inefficient to avoid the death of one patient who had long lasting lesions in the liver with obstruction of the biliary system and portal hypertension¹¹. The same failure was observed in two of the Meneghelli patients treated with albendazole²⁰.

Surgical treatment was effective in removing the metacestode from the base of the mesentery and the patient was reported cured (case 4 of Meneghelli)²¹. In 5 of the 20 Brazilian PHD cases, their fate was unreported. In our experience and in the study of the results reported by others, surgical treatment should be recommended when the metacestode is small, localized and has not responded to medical treatment. Radical cures

may be expected in these cases, but probably not when the PHD involves a large portion of the liver or other organs. However, more observations for prolonged periods of time are necessary to assess the chances of permanent improvement or cure in each case.

When the biliary system is involved, or cirrhosis has developed with splenomegaly and esophageal varices, surgery has accelerated the death of the patient due to complications. However, if the itching due to the jaundice is very troublesome, drainage of the biliary tract is usually required.

Surgery has been used successfully as a palliative mean of diminishing the effects of large numbers of cysts, which interfere with normal physiological activity of the stomach and other organs, by removing as many cysts from the mesenteries or those emerging from organs as possible. In addition, the absence of intraperitoneal masses allows the patient to sleep more comfortably (D'Alessandro, unpublished).

In summary, medical treatment with albendazole has been used with good results although really it is too early to assess long lasting cures. However, albendazole should be used and evaluated on the patient's well being. In general, *Ev* seems to be less aggressive than *E. multilocularis* and we know of asymptomatic patients who had the infection for several decades.

The epidemiological information obtained from the Brazilian cases is valuable. The Acre patients not only were familiar with pacas but also had seen parasitic vesicles in their abdomen. As in Colombia, the viscera of the pacas are fed to dogs. The conditions in Acre, at least, seem favorable for studies of transmission of *Ev* due to the number of infected persons reported and unreported²⁰. In this place it has not been established how the pacas get the infection, but Acre is within the geographical distribution of the bush dog. In 1986⁹ it was reported 13 specific places where the bush dog had been seen in Brasil including the states of Minas Gerais, Mato Grosso, Santa Catalina and São Paulo, and these places were shown in a map (Figure 9). Also unknown is the frequency of infection in pacas, in domestic dogs and in humans (which could be assessed by using the latest available serological tests).



Figure 9 - Map 1 showing the distribution of the bush dog, *Speothos venaticus*. From Peter Druwa, reference 9. Courtesy of the author and the Editor of the journal.

Figure 10 - Map 2 showing the distribution of pacas, *Cuniculus paca* and agoutis, *Dasyprocta* spp, from different sources.

In Colombia, people from the enzootic area could not provide information on the presence of the bush dog until a photo of the animal was presented. Then most people recognized the animal. The local name in the Colombian Oriental plains is "zorro guache" and in Brazil the local name is "cachorro do mato vinagre"³.

A survey of a limited random sample of adult pacas (weighing more than 10kg) should be sufficient to assess the prevalence of Ev infection, considering that the frequency in Colombia was 30% in adult animals, 16% in subadults and none in the juveniles weighing less than 5.5kg⁸.

Although Ev infects humans and animals more frequently than *E. oligarthrus* the geographical distribution of wild cats is wider than that of the bush dog⁸. Therefore, the presence of PHD outside the range of the bush dog may be due to *E. oligarthrus*. However, it has been postulated that animals other than the bush dog and pacas may be involved in the transmission of Ev⁶.

Actually, both possibilities may play a role. To answer this question the collaboration of

zoos is required by the treatment with praziquantel of canids/felids that have been capture in the wild as well as those which has been in captivity, in particular for a short time. Long lasting captivity may have an effect on some parasites which are eliminated spontaneously.

It is important to determine the species involved in a case of PHD because, apparently, Ev is susceptible to medical treatment with albendazole although, as stated earlier, long term observations of reported successes require prolonged periods of observation. A number of reports are encouraging, at least in regard to the improvement of the patient's health.

RESUMO

Este trabalho descreve cinco novos casos humanos brasileiros de doença hidática policística, causada por Echinococcus vogeli e faz uma revisão de casos anteriores relatados no Brasil, incluindo um relato sobre E. oligarthrus (num total de 20) e alguns aspectos epidemiológicos desta doença, que já não é mais uma curiosidade mas, sem dúvida,

um problema de difícil manejo do ponto de vista médico. Sua presença pode ocorrer em todas as áreas rurais do Novo Mundo, onde o homem ainda não eliminou os felinos e caninos selvagens, cachorro do mato, paca, cutia e outros roedores selvagens.

Palavras-chaves: Doença hidática policística. *Echinococcus vogeli*. Brasil.

ACKNOWLEDGMENTS

We thank Dr. Robert L Rausch (Department of Comparative Medicine, University of Washington, Seattle, WA.) and Dr. M Dale Little (Department of Tropical Medicine, Tulane University School of Public Health and Tropical Medicine, New Orleans, LA.) for reviewing the manuscript and offering useful suggestions.

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