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ORIGINAL PAPER

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Human cystic echinococcosis in the West Bank of Palestine: surgical incidence and seroepidemiological study

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Abstract The surgical incidence of cystic echinococcosis (CE) due to *Echinococcus granulosus* was investigated in hospitals of the West Bank, Palestinian Authority between January 1990 and December 1997. Serum samples from school-children in Yata town, which showed the highest surgical incidence, were tested for anti-hydatid antibodies. A total of 390 surgically confirmed cases were recorded throughout the 8-year period, with an overall mean annual surgical incidence (MASI) of 3.1 per 100,000. A high MASI of 4.9, 5.0 and 5.1 per 100,000 was found in Hebron, Jericho and Bethlehem Governorates, respectively. Yata town, Hebron governorate, showed the highest MASI, at 16.8 per 100,000. The highest incidence was found in age groups 11–20 and 21–30 years, at 27.4% and 21.5% of the total number of cases. While there was no significant gender difference in the number of cases in the age groups of 20 years or less, the male to female case ratio was 1:3.2–4.1 in the older age groups. The liver was the most common site of hydatid cysts in 69.9% of cases. Lung cysts were predominant in younger age groups (20 years or less). The seropositivity for CE in the school-children of Yata was 2.4% and 2.1% using enzyme-linked immunosorbent assay and the indirect haemagglutination test, respectively. CE is a significant endemic disease throughout the West Bank. The disease is acquired early in life and is more prevalent among females than males. Behaviour and life-style favour the spread of the disease.

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Introduction

Cystic echinococcosis (CE), caused by infection with the larval stage (hydatid) of the small dog tapeworm *Echinococcus granulosus*, is a chronic zoonotic disease of major public-health importance. The parasite is transmitted cyclically between the dog as definitive host and sheep and other livestock as intermediate hosts. The disease has a global distribution in most regions where sheep-raising is a major industry. The incidence of the disease is particularly high in rural areas, where there is increased contact between man and dog. Human CE is often considered an occupational public-health problem for sheep farmers, ranchers and shepherds in endemic areas (Cohen et al. 1997). The disease is endemic in many countries throughout the world, including the Middle East (Schantz et al. 1995; Abdel-Hafez and Kamhawi 1997). Within the Middle East, CE is a significant public-health problem in several countries, including Jordan, Iraq, Lebanon, Arabian Peninsula and North African countries (Abdel-Hafez and Kamhawi 1997; Dar and Alkarmi 1997; Ibrahim and Gusbi 1997; Kachani et al. 1997; Ouhelli et al. 1997; Shambesh et al. 1997). In Israel, a major focus of endemicity is found in the northern Arab villages of Yirka and Tamra (Goldsmith et al. 1991; Nahmias et al. 1993, 1994; Furth et al. 1994; El-On et al. 1997; Hoida et al. 1998).

The most commonly used index of human infection is the annual incidence of hospital surgical cases. Despite the limitation of this method, data on annual rates proved to be of great value for defining public-health importance, providing new information about the epidemiology of the disease, and documenting its spread in different localities and among various groups at risk.

Several studies have dealt with the mean annual surgical incidence (MASI) in countries that surround the West Bank. In Jordan, Kamhawi (1995) reported on the MASI of CE between 1985 and 1994. Overall the MASI was 2.9/100,000, being highest at 8.2/100,000 in the Karak rural population. Women were more affected,

and a significant percentage of the cases were children below 10 years of age. In Palestine and Israel, the earliest records on human incidence of CE give 1/100,000 during the period 1922–1935, and the incidence has increased over the years, reaching as high as 5/100,000 during 1959 (Yarrow et al. 1991). In northern Israel, a resurgence of CE was noted among Arab and Druze communities beginning in early 1970. In these communities, the MASI recorded between 1980 and 1989 was five-fold that recorded in 1960–1969 (Goldsmith et al. 1991). In the West Bank, surgical records from the Al-Maqased Hospital in Jerusalem estimated a MASI of 1.76/100,000 in 1995 (see Abdel-Hafez and Kamhawi 1997).

Seroprevalence studies are important in identifying foci of active CE infections, particularly when coupled with ultrasound methods (Cohen et al. 1997; Craig 1997; Shambesh et al. 1999). In Jordan, the seropositivity rate in the general population and among school-children was 2.4%–2.8% (Abu-Shehada 1993; Moosa and Abdel-Hafez 1994), but higher seropositivity rates, of 4.4% and 5.9%, were noted among shepherds and at certain hot foci in Jordan (Abdel-Hafez and Kamhawi 1995).

This investigation reports on the surgical incidence of CE in various general hospitals throughout the West Bank of the Palestinian Authority over a period of 8 years (January 1990-December 1997). Anti-hydatid seropositivity was determined in school children from Yata town, which showed the highest surgical incidence for the disease. The life-styles and practices of the Yata population that led to such a high surgical incidence were investigated.

Materials and methods

Study area and community

The Palestinian Authority oversees the Arab Palestinian communities in the West Bank and Gaza Strip, with a total population of 2.9 million, of which 1.9 million reside in the West Bank. Many Jewish settlements exist in the West Bank, but their population forms part of the Israeli community in both life-style and civil services. Consequently, these settlements were not included in the present study. The West Bank is divided into nine governorates, ranging in population size from 72,000 (Qalqiliah Governorate) to 406,000 (Hebron Governorate). Each governorate has a central town around which satellite villages are scattered. Most rural populations raise sheep and goats in close proximity to their households. According to the 1996 census, 270,825 sheep, 1,271 cattle, 1,146 camels and 3,707 donkeys were reported in Hebron Governorate. This represented 32.8% of the total livestock count in the West Bank (Ministry of Agriculture-Annual Report 1996). Traditional animal husbandry and the practice of slaughtering animals at home are very common.

Surgical incidence

All nine West Bank governorates are served by seven public hospitals (Rafeedia, Jenin, Tulkarem, Ramalla, Alia, Jericho, Al-Hussein) and five private general hospitals (Al-Itihad, Saint Lukes, Al-Ahli, Al-Maqased, Augesta, Vectoria). In this study, all records of surgery performed in these hospitals throughout the period January 1990–December 1997 were inspected for confirmed surgical cases of CE. Data regarding place of residence, age, gender, previous cyst location, number and size, as well as the previous history of the disease, were recorded. In some instances, specific data were not available in the files examined and these records were not included. The compiled data were analysed using an SPSS software program.

Serological testing

Yata town in Hebron Governorate has a population of 30,800 (1997 census). The town is in a rural area and most of its inhabitants raise domestic animals, including sheep and cattle. Stray dogs are abundant in the vicinity of the town, although their number was not estimated. As the population of Yata town showed the highest surgical incidence for the disease, the seropositivity rates for CE in school-children in the town were investigated. Blood samples were collected from 190 female students from Riqah Girls School, and 137 male students (ages 7–14) from Yata Primary School. A specially designed questionnaire was used to collect demographic data covering the practices and life-styles of Yata inhabitants, as pertaining to CE, through questioning 203 female students. For serology, 2.5 ml blood samples were collected from each subject by venipuncture. Sera were separated and stored at –20 °C until used. Sera samples were assayed for the presence of anti *E. granulosus* lgG antibodies using enzyme-linked immunosorbent assay (ELISA) and indirect haemagglutination (IHA) tests with crude sheep hydatid fluid as the target antigen. ELISA and IHA were performed as previously described (Moosa and Abdel-Hafez 1994).

Results

Annual surgical incidence of CE cases

A total of 390 surgically confirmed CE cases were found in the records of surgical hospitals of the West Bank of the Palestinian Authority for an 8-year period 1990–1997. The number of cases per year ranged between 29 and 63. On the basis of a population estimate of the West Bank in 1997 (1.873 million) and an estimated growth rate of 4.9% (Microsoft Encarta 1998), MASI ranged between 2.2 and 3.7 per 100,000 (1990 and 1995, respectively) (Fig. 1). The overall MASI was 3.1 per 100,000 (Table 1). The highest MASI was recorded in the Hebron, Jericho and Bethlehem Governorates (4.9–5.1 per 100,000), while the lowest was in the Qalqilia and Jerusalem Governorates (0.9–1.0 per 100,000). The number of CE surgical cases from Hebron Governorate

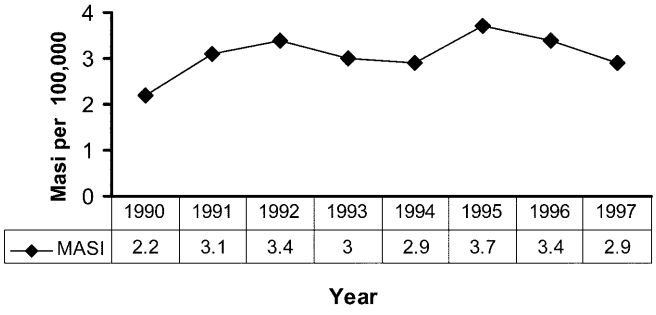

Fig. 1 Mean annual surgical incidence (MASI) of cystic echinococcosis per 100,000 in West Bank, Palestinian Authority (1990–1997)

Table 1 Number of cases of cystic echinococcosis (*CE*) and mean annual surgical incidence (*MASI*) in various governorates of the West Bank of the Palestinian Authority (1990–1997)

Governorate	Mean population ($\times 10^3$)[a]	Total no. of cases[b]	Mean no. of cases/year	MASI per 100,000
Hebron	342	140	17.5	5.1
Jericho	28	11	1.4	5.0
Bethlehem	116	45	5.6	4.9
Jenin	202	59	7.4	3.7
Ramallah	180	51	6.4	3.6
Nablus	261	37	4.6	1.8
Tulkarm	113	15	1.9	1.7
Qalqilia	61	5	0.6	1.0
Jerusalem	277	20	2.5	0.9
Unknown	–	7	0.9	–
Total	1582	390	48.8	3.1

[a] The mean population of each governorate was calculated on the basis of the 1997 population estimate of 1.873 million, taking into consideration a 4.9% annual growth rate

[b] Total number of cases for the 8-year period (1990–1997). The number of cases per year ranged between 29–63

was consistently high throughout the 8-year period and comprised 36.6% of all cases. Out of the 140 cases reported in Hebron Governorate, 35 (25%) were patients living in the town of Yata. This town had an estimated mean population of 26,000 during the 8-year period. Thus, the MASI of CE in this town was calculated at 16.8/100,000.

Age and sex distribution of surgical cases

Age groups 11–20 and 21–30 were the most affected groups and constituted 27.4% and 21.5% respectively of the total number of cases (Table 2). A significant percentage (12.1%) of the cases were 10 years of age or younger. Females constituted 62.3% of the total number of cases. The overall gender difference was statistically significant at $P < 0.005$. There was no significant gender difference in the number of cases aged below 20 years (male to female case ratio 1:1–1.1). In contrast, significantly more female than male cases were recorded throughout the age groups between 31 and 60 years, with male to female case ratio being 1:3.2–4.1 (Table 2).

Cyst location, size and recurrence

The liver was the most common site of CE infection, accounting for 58.2% of the cases as a single site for hydatid cysts and in 11.7% of these cases were in combination with other sites (Table 3). The lung was the second most frequently involved site of infection (20.8% as a single site and 5.1% in combination with the liver). The number of patients with lungs as the single site of infection exceeded by 4× the number of those who had concurrent lung and liver hydatid cysts (Table 3). Lung cyst involvement was seen in younger age groups. Out of the total lung involvement in 101 patients, 58.4% were in the age group 20 years or less. Single cysts comprised 42.3% of the total number of cases and as many as 72.5% of the cases had only one surgical intervention for the extraction of hydatid cysts. In 69.7% of the cases the cyst diameter exceeded 5 cm. The largest recorded cyst was 20×20×20 cm.

Serodiagnosis

Serodiagnosis of school-children (ages 7–14) from Yata town, Hebron Governorate using ELISA and IHA revealed an overall seropositivity of 2.4% and 2.1%, respectively (Tables 4). Males showed a significantly higher seropositivity rate than females in both tests. Two of the weakly ELISA-positive samples (nos. 173 and 351) were actually negative using IHA (Table 4). One of the seropositive samples (no. 360) was for a confirmed case of CE. This case scored a very high positivity in both tests.

Table 2 Age and sex distribution of surgically confirmed CE cases recorded in the West Bank of the Palestinian Authority (1990–1997)

Age group (years)	No. of cases			% of total	Male: female ratio
	Males	Females	Both		
< 10	23	24	47	12.1	1:1.0
11–20	52	55	107	27.4	1:1.1
21–30	30	54	84	21.5	1:1.8
31–40	9	37	46	11.8	1:4.1
41–50	6	19	25	6.4	1:3.2
51–60	8	33	41	10.5	1:4.1
> 60	19	21	40	10.3	1:1.1
Total	147	243	390	100.0	1:1.7

Table 3 Cyst locations in surgically confirmed CE cases from the West Bank of the Palestinian Authority (1990–1997)

Site	No. of cases	% of total
Liver alone	227	58.2
Lung alone[a]	81	20.8
Liver and lung[a]	20	5.1
Liver and kidney	11	2.8
Liver and other organs	15	3.8
Spleen	4	1.0
Brain	4	1.0
Kidney	3	0.8
Other organs[b]	6	1.5
Unspecified	19	4.9
Total	390	100.0

[a] Out of the total number of 101 cases with lung cysts, 28 (27.7%) and 31 (30.7%) were found in patients who were < 10 and 10–20 years of age, respectively
[b] These sites included the ovary (1), spinal chord (1), thyroid (1), peritoneal cavity (2), and diaphragm (1)

Table 4 Correlation between ELISA (for IgG) results and IHA titres for serum samples of students from Yata town (Hebron Governorate, West Bank of the Palestinian Authority). The total number of samples tested was 327 (190 females and 137 males)

Sample no.	Sex	ELISA[a]	IHA[b]
1	F	+	1:64 + (+)
173	F	+	1:32 (–)
233	F	+ +	1:6,400 (+ + + +)
282	M	+ +	1:800 (+ +)
288	M	+ +	1:64 (+)
337	M	+	1:400 (+ +)
349	M	+ +	1:128 (+)
351	M	+	1:32 (–)
360 [c]	M	+ + + +	1:12,800 (+ + + +)
Total and (%) positive			
	F	3 (1.6%)2 (1.1%)	
	M	6 (4.4%)	5 (3.6%)
	Both	9 (2.4%)	7 (2.1%)

[a] The ELISA score was based on the mean × optical density (OD) of at least six negative control samples tested in each plate and used as a cut-off point (COP). Scoring of test samples was as follows: (+) for OD > COP- < 2×COP; (+ +) for OD > 2×COP- < 3 XCOP; (+ + +) for OD > 3×COP- < 4×COP; (+ + + +) for OD > 4COP
[b] The COP titre for IHA was 1:64. Any titre less than 1:32 was scored as (–). Positive titres were scored as follows: (+) for titres of 1:64–1:200; (+ +) for titres 1:400–1:800; (+ + +) for titres 1:1,600–1:3,200; (+ + + +) for titres 6,400 and above
[c] This sample was from a proven CE case from Yata town community

Five of the seropositive samples were from members of families with a long history of animal-raising, especially sheep. Three of them had sheep-dogs and all reported seeing stray dogs in their residential areas. Three reported that their house water-supply was from rainwater collected in wells. None of the seropositive subjects reported disease occurrence among any of their family members.

Behaviour and life styles

Out of 208 female students who were interviewed to determine the behaviour, practices and life-styles of their families, 60.4% came from families that raised domestic animals. Stray dogs were reported in the vicinity of the town by 75.1% of the students, and 29.4% of the families had one or more dogs within their households (Table 5). Dead and condemned offal not suitable for human consumption was thrown in the open by 80.3% of the families. With respect to water supply, 60.0% of the student families depended on rainwater collected in wells as the main source of their water supply and 5.2% drink water collected from nearby springs (Table 5).

Discussion

This study represents the first comprehensive investigation of the surgical incidence of CE in the West Bank of the Palestinian Authority. The overall MASI of 3.1/100.000, as reported here, represents an under-estimation of the actual incidence for several reasons. First, this incidence does not take into account patients who underwent surgery for CE in both Israel and Jordan, where some Palestinians are referred for surgical intervention. Secondly, not all patients infected with CE undergo surgery; there are several asymptomatic cases for each surgically confirmed case. In this respect, seroepidemiology and imaging techniques using ultrasound are better indicators of the actual rate of incidence (Craig 1997). Thirdly, hospital records may not account for all cases of surgery for CE, due to poor or limited filing systems.

In spite of the above-mentioned limitations, CE appears to be a significant disease of stable endemicity in the West Bank. The MASI was consistently high during the 8-year period of study, in the range 2.2–3.7/100.000 (Fig. 1). This incidence contrasts with a lower MASI of 1.76/100.000, reported earlier for cases recorded at Al-Maqased Hospital in Jerusalem (Saifi, personal com-

Table 5 Percentage of female students who answered questions pertaining to behaviour and life style at Yata town, Hebron Governorate, West Bank of the Palestinian Authority (1997 survey). Percentages were based on a questionnaire completed after interviewing 208 female students

Questions on behaviour or life style	Percentage
1. Family raise livestock in household	60.4
2. Family own dogs	29.4
3. Stray dogs seen nearby residence	75.1
4. Family throw dead animals in the open	80.3
5. Family burn dead animals	8.3
6. Family bury dead animals	11.4
7. Family drink water from municipality pipes	34.8
8. Family drink water from wells containing rain water	60.0
9. Family drink water from springs	5.2

munication; see Abdel-Hafez and Kamhawi 1997). The incidence reported here is similar to surgical incidences reported in nearby countries, such as Jordan (2.9/100,000), Lebanon (3.6/100,00) and Kuwait (3.6/100,000) (Schwabe and Abou-Daoud 1961; Shweiki et al. 1990; Kamhawi 1995). Residents of Hebron, Jericho, Bethlehem, Jenin and Ramallah are considered at higher risk of contracting CE than those of other localities (Table 1). The town of Yata in Hebron Governorate appears to be the hottest spot for CE, with a MASI of 16.8/100,000.

In the West Bank, CE is contracted at an early age, below 10 years. The greatest number of cases that warranted surgical intervention appeared in the age-groups 11–30 years (Table 2). As cysts of *E. granulosus* are known to develop slowly over several years, manifestation of symptomatic disease would be expected to occur in these age groups. At young ages, no significant difference in surgical incidence was noted between males and females. The observed differences of CE among cases below 20 years can be attributed to the different occupational roles played by males and females in Palestinian rural communities (Table 2). Women appear to be more exposed to the eggs of the parasite, either through closer contact with dogs, or through their domestic and agricultural practices. The trend toward higher surgical incidence among women is consistent with reports from other Arab countries, including Jordan (Kamhawi 1995; Abdel-Hafez and Kamhawi 1997) and North African countries (El Idrissi et al. 1997; Shambesh et al. 1997).

Further evidence of early exposure to parasite comes from the significant number of lung cases detected, particularly in patients less than 20 years of age (Tables 3). It appears that patients contracting the infection during childhood are at higher risk of lung than liver involvement. The present data reinforce this observation, which was also noted by Chaouachi et al. (1989) among Tunisian children. The finding that 69.7% of cases have cysts greater than 5 cm in diameter is an indicator of the slow growth of the parasite to reach symptomatic size warranting surgical intervention. Late diagnosis and/or lack of awareness of the possible complications associated with the disease are also contributing factors to cysts of large size. Evidence in support of such an assumption can be deduced from the findings of three reported cases with trauma, due to rupturing of cysts, that resulted in anaphylactic shock, and the finding of a patient with several hundred cysts distributed within the visceral cavity. Recurrent cases constituted a significant proportion of the surgically confirmed cases. The limited use of imaging techniques, including CT scan and computer axial tomography, or misinterpretation of the results of such techniques may also account for the late detection of hydatid cysts.

A seropositivity rate of 2.1%–2.4% among 7- to 14-year-old school-children from Yata town was obtained using ELISA and IHA, (Table 4). This rate is similar to the overall seropositivity rate reported in Jordan (Moosa and Abdel-Hafez 1994), but is significantly lower than rates reported in highly endemic foci in northern Jordan, where a seroprevalence rate of 5.9% was reported (Abdel-Hafez and Kamhawi 1997). The seroprevalence rate in Yata town is higher than that reported in Arab/Druze communities of Yirka town in northern Israel, which was estimated to be 1.6% (Nahmias et al. 1993). In contrast, a much higher seroprevalence rate (10.5%) in the age group 0–14 years was detected in Libya (Shambesh et al. 1999).

The fact that the seropositivity rate among male students exceeded 2× that of females in Yata is difficult to explain at present. This is particularly important in light of the contrasting observations of no significant differences in the overall surgical incidence between males and females in the young age-groups (less than 20 years), as indicated above (Table 3). Moreover, the seropositivity data contrast with findings by Abdel-Hafez (unpublished observations) in Jordan, in which no significant difference in the seroprevalence was noted in similar age groups.

The town of Yata provides an excellent focus for the study of the epidemiology and transmission dynamics, including prevalence and incidence rates in humans, through surgical incidence, serodiagnosis and imaging techniques, as well as demographic parameters, socioeconomic criteria, and prevalence in livestock (intermediate hosts) and dogs (final host). It is noteworthy that certain families in this town have as many as 18 members, as polygamy is common. Such a situation is likely to be associated with poor hygiene, due to over-crowding. The abundance of stray dogs in the vicinity of residential areas (Table 5) and poor hygiene go hand-in-hand to explain the high seroprevalence and surgical incidence rates. In this context, determination of the infection rates of stray dogs with *E. granulosus,* either by necropsy or through detection of *E. granulosus* coproantigens in dog fecal samples, is of the utmost importance.

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