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The changing incidence of human hydatid disease in England and Wales

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SUMMARY

The incidence of hospital-diagnosed human hydatid disease acquired in the UK was estimated from a survey based on Hospital Activity Analysis data for the period 1974–83. The average annual incidence in Wales was 0·4 per 100000 population compared with 0·02 per 100000 in England. Within Wales, Powys, and particularly Brecknock, had the highest incidence (7 per 100000 per year). Compared with the period 1953–62, the average annual incidence for Wales fell by half (from 0·8 to 0·4 per 100000 per year), but in Powys the incidence did not decline, and in Brecknock and Montgomery there was a marginal increase. In comparison with 1953–62, the age-specific incidence in Wales and Powys decreased in each age group with the notable exception of children < 15 years of age. This finding emphasizes that transmission of *Echinococcus granulosus* to humans is still occurring at hyper-endemic levels in parts of England and Wales and that control efforts should be intensified.

INTRODUCTION

Human hydatid disease is caused by infestation with the larval stage of the dog tapeworm *Echinococcus granulosus*, and is acquired by ingesting the eggs of the tapeworm which are excreted in the faeces of the dog (Schantz, 1982). The important intermediate host for *E. granulosus* in Britain is sheep, and the sheep-farming areas of Mid-Wales have been known for many years to be a focus of endemic hydatid disease (Howell, 1940; Walters, 1977; Howells & Taylor, 1980). A veterinary control programme organized by the State Veterinary Service and supported by the Welsh Office was set up in 1983 in South Powys (Walters, 1986). As part of this programme an accurate measure of the incidence of human hydatid disease in different areas of England and Wales was required to determine the size of the public health problem and to provide a baseline from which to monitor the effect of control measures. The use of hospital data to measure incidence was recommended for population surveillance by the World Health Organisation (Eckert, Gemmell & Soulsby, 1981), and has been used to monitor the successful control programme in Tasmania (Beard, 1978). We used hospital-diagnosed cases to estimate the incidence of UK-acquired human hydatid disease in 1974–83 and compared the results with a similar study carried out for the period 1953–62 (Thomas, 1966).

METHODS

Data on hospital admissions were sought through Hospital Activity Analysis (HHA) with ICD code 122 to identify possible cases. Preliminary discussions with Regional Health Authorities suggested that HAA date prior to 1974 would be seriously incomplete. Between the 14 Regional Health Authorities and the Welsh Office there were seven different procedures that had to be followed eventually to obtain access to medical records. In all cases, permission was sought individually from clinicians to review medical records of indigenous patients who were first diagnosed as having hydatid disease after 1974.

Two studies were undertaken to try to estimate the extent of under-reporting in HAA. First, all hospital histopathologists in Wales and the West Midlands where cases of hydatid disease were known to have been admitted, and others in hospitals in other regions where two or more cases were identified, were asked if they could provide data on confirmed cases. Secondly, laboratory data derived from the serology reference service at Liverpool University and Cardiff PHL were compared with HAA data.

To investigate the possible sources of infection in English resident cases (excluding Herefordshire, which was known to be a high endemic area) the general practitioners (GP) of patients < 50 years of age were contacted. A case-control study was conducted to test the hypothesis that English cases were more likely to have lived in or visited Wales before diagnosis of hydatid disease. GPs were asked to provide the names and addresses of three controls for each case from their practice registers matched for age (± 5 years) and sex. In practices with an age/sex register, controls were the next three patients to the index case who were not relatives, and in other practices, controls were selected from practice records as the nearest three patients to the index case fulfilling the matching criteria.

RESULTS

HAA data

Eighty-eight percent (572/646) of HAA-identified patients in England and 98% (185/188) in Wales were traced (Table 1). Twenty-four per cent (138/572) of the English HAA cases and 8% (14/185) of Welsh HAA cases were incorrectly coded and were not hydatid disease. A total of 201 cases, mainly foreign nationals, definitely acquired hydatid disease outside the UK. From the review of medical records and contact with general practitioners, a further 13 cases were found to have lived overseas in endemic hydatid areas, and we considered that to be their probable source of infection. Thus there were 206 new cases of hydatic disease possibly acquired in the UK, 104 in Wales and 102 from English Health Authorities.

Survey of histopathologists and laboratory reports

Thirty-four histopathologists were contacted, and 19 provided data on 34 patients; 30 of these were known from HAA, and one new post-1973 UK-acquired case was identified. From laboratory data, 7 post-1973 UK-acquired cases were identified and only one was known from HAA. Two other cases were identified by

Table 1. *Results of Hospital Activity Analysis survey of human hydatid disease*

Health Authority*	No. of patients	Successfully traced	New cases	First diagnosed pre-1974	Overseas	Miscoded
			England			
Northern	15	15	2	1	3	6
Yorkshire	49	45	7	2	9	12
Trent	26	24	7	2	6	7
E. Anglia	10	10	1	—	6	4
N.W. Thames	72	62	4	3	28	17
N.E. Thames	127	115	2	5	74	17
S.E. Thames	112	76	—	3	39	29
S.W. Thames	16	14	2	1	7	3
Wessex	18	18	2	1	3	4
Oxford	15	15	5	—	6	1
South Western	14	14	7	4	1	2
W. Midlands	115	115	47	6	19	19
Mersey	14	13	4	—	3	4
North Western	43	41	12	—	7	13
England Total	646	572 (88%)	102 (18%)	28 (5%)	211 (37%)	138 (24%)
			Wales			
E. Dyfed	14	14	5	4	—	3
W. Glamorgan	19	19	9	3	2	2
Clwyd	13	13	3	4	—	1
Powys	11	11	11	—	—	—
S. Pembs	1	1	—	—	—	1
Mid-Glamorgan	25	25	19	3	—	1
Gwent	40	40	23	10	—	2
S. Glamorgan	52	52	29	4	1	3
Gwynedd	13	10	5	—	—	1
Wales Total	188	185 (98%)	104 (57%)	28 (15%)	3 (2%)	14 (8%)

* Health Authority in which diagnosis was first made, not necessarily the HA of residence.

clinicians during the course of the study. Therefore, in total, data from 215 patients fulfilling the study criteria became available for analysis.

Incidence of human hydatid disease

Powys, and particularly Brecknock, had the highest incidence (Table 2). Outside Powys, districts bordering on Brecknock, namely Dinefwr, Blaenau Gwent, Monmouth and South Herefordshire (Table 3) had relatively high incidence rates.

The incidences estimated in our study were compared with data collected by Thomas (1966) for the period 1953–62, from which we calculated rates based upon current geographical boundaries and 1961 Census figures. For Wales as a whole, the average annual incidence fell by half, but in Powys there was no decline (Table 4). In Brecknock and Montgomery, the incidence marginally increased.

The age-specific incidence of human hydatid disease in Wales as a whole shows

Table 2. *Incidence of hospital-diagnosed human hydatid disease in Wales by district,* 1974–83*

District	No. cases	Incidence† /100000/year
Wales	115	0·4
Clwyd	2	0·05
Alun and Deeside	1	0·1
Colwyn	—	—
Delyn	—	—
Glyndwr	1	0·3
Rhuddlan	—	—
Wrexham Maelor	—	—
Dyfed	10	0·3
Carmarthen	3	0·6
Ceredigion	2	0·4
Dinefwr	3	0·8
Llanelli	2	0·3
Preseli	—	—
S. Pembrokeshire	—	—
Gwent	15	0·3
Blaenau Gwent	6	0·8
Islwyn	1	0·1
Monmouth	5	0·7
Newport	3	0·2
Torfaen	—	—
Gwynedd	6	0·3
Aberconwy	3	0·6
Arfon	2	0·4
Dwyfor	—	—
Meirionnydd	—	—
Ynys Mon	1	0·1
Mid-Glamorgan	25	0·5
Cynon Valley	1	0·1
Merthyr Tydfil	2	0·3
Ogwr	9	0·7
Rhondda	4	0·5
Rhymney Valley	5	0·5
Taff-Ely	4	0·4
Powys	43	4·0
Brecknock	28	7·1
Montgomery	5	1·0
Radnor	10	4·8
South glamorgan	7	0·2
Cardiff	4	0·1
Vale of Glamorgan	3	0·3
West Glamorgan	7	0·2
Afan	1	0·2
Lliw Valley	1	0·2
Neath	1	0·1
Swansea	4	0·2

* By area of residence.

† 1981 Census figures, usually resident population.

Table 3. *Incidence of hospital-diagnosed hydatid disease in the West Midlands Health Authority Region by county and district,* 1974-83*

County/District	No. cases	Incidence/ 100000/year†
England	100	0·02
Hereford and Worcester	11	0·2
Bromsgrove	—	—
Hereford	1	0·2
Leominster	—	—
Malvern Hills	2	0·2
Redditch	—	—
South Herefordshire	5	1·1
Worcester	2	0·3
Wychavon	—	—
Wyre Forest	1	0·1
Shropshire	1	0·02
Staffordshire	6	0.06
Warwickshire	—	—
West midlands	19	0·07
Birmingham	10	0·1
Coventry	—	—
Dudley	3	0·1
Sandwell	—	—
Solihull	3	0·1
Walsall	—	—
Wolverhampton	3	0·1

* By area of residence.
† 1981 Census figures, usually resident population.

Table 4. *Average annual incidence of hospital-diagnosed human hydatid disease in Powys**

District	Incidence/ 100000/year (1953–62)	Incidence/ 100000/year (1974–83)
Brecknock	5·8	7·1
Montgomery	0·7	1·0
Radnor	4·9	4.8
Powys	3·7	4·0
Wales	0·8	0·4

* Based on 1961 and 1981 Census population figures.

a general trend of increasing incidence with age, although this trend was more evident in the 1953–62 data (Table 5). Compared with 1953–62, the age-specific incidence for Wales and for Powys has decreased in each age group with the exception of children < 15 years of age.

Sources of infection in English cases

Of the 92 UK-acquired English cases living outside Herefordshire, three cases had only recently moved to England from sheep farms in Mid-Wales. One other woman visited Welsh farms regularly. Three patients had been brought up in Mid- or South Wales, one on a sheep farm. Three may have acquired infection from

Table 5. *Incidence/100000/year of hospital-diagnosed human hydatid disease by age**

Age range (years)	Wales 1953–62 (cases)	Wales 1974–83 (cases)	Powys 1953–62 (cases)	Powys 1974–83 (cases)
0–4	0·1 (2)	0·2 (3)	1·1 (1)	4·6 (3)
5–14	0·3 (12)	0·4 (15)	2·2 (4)	4·4 (7)
15–29	0·6 (29)	0·3 (20)	5·1 (11)	4·9 (11)
30–44	0·6 (30)	0·2 (13)	3·5 (8)	2·8 (6)
45–64	1·4 (93)	0·6 (40)	5·2 (16)	4·6 (12)
⩾ 65	1·4 (45)	0·5 (23)	2·5 (4)	2·0 (4)
Not known	— (4)	— (1)	— (-)	— (-)
Total	0·8 (215)	0·4 (115)	3·7 (44)	4·0 (43)

* Based on 1961 and 1981 Census population figures.

dogs bred in Wales. These were a sheep farmer and his 28-year-old daughter, who developed hydatid disease within 2 years of each other after buying a sheepdog from Mid-Wales, and a man whose dog was proved to harbour the tapeworm. One case had lived most of his life in Herefordshire. Another had frequently visited grandparents on a farm in North Wales in his childhood. One individual had worked in forestry in Wales as well as on a sheep farm in England.

Of the 39 English patients less than 50 years of age, 2 had died, 10 were not registered with a GP and one was untraceable. The remaining 26 were included in the case-control study and results were obtained for 24 and their 61 matched controls. Five cases had previously lived in Wales or Herefordshire (Brecon, Hereford, Painscastle, Rhymney and Sennybridge) as had two controls (Milford Haven and Hereford). The relative risk for ever living in Wales or Herefordshire was 10·1 (95% confidence intervals 1·1–88·2). A history of holidays in Wales and dog ownership were not significantly different between cases and controls, although three cases, but no controls, had been on holiday in the Brecon area. Three of the 24 cases had never owned a dog.

DISCUSSION

The incidence estimates for human hydatid disease reported in this study were based primarily upon HAA data. Since treatment for hydatid disease will almost always involve hospital admission, HAA potentially should provide a complete list of treated patients. There may be others, however, particularly elderly patients with inactive calcified cysts indentified by general practitioners, who are not investigated in hospital. Also, an unknown number of people will have asymptomatic disease. Hospital data therefore cannot give a complete picture of infection with *E. granulosus* in the whole population. Despite these deficiencies, WHO (Eckert, Gemmell & Soulsby, 1981) recommended that hospital cases represent one of the most useful and practicable indicators of the success of control measures. In particular, hospital data will indicate foci of infection where control measures should be directed.

The major deficiency of the HAA system discovered in our study was the 24%

diagnostic code error rate in English regions. It is clear that HAA cannot be used as a primary source of incidence data without confirming the diagnostic coding with the patients' records. Given the error rate of the English HAA, we were concerned about the degree of under-recording of hydatid disease. Our survey of histopathologists and laboratory reports revealed 7 new cases out of 39 patients identified. Though these data are limited they do suggest that HAA was something like 18% incomplete, and our data will therefore underestimate the true incidence of hospital-diagnosed hydatid disease.

The study confirmed that South Powys is the main focus of human hydatid disease in the country (Walters, 1978). It also revealed that human hydatid disease has virtually disappeared from North Wales. Thomas (1966) recorded 39 of 215 Welsh cases living in Caernarvonshire, Anglesey, Merioneth, Denbighshire and Flintshire, compared with only 8 of 115 cases in Gwynedd and Clywd in our survey. These data on human infection are in agreement with veterinary data, which showed a high prevalence of infection in farm dogs in Mid-Wales but virtually no infection in dogs in North Wales (Edwards, Hackett & Herbert, 1979; Stallbaumer, 1985). No satisfactory explanation has been discovered for this finding.

The most important finding was that the incidence in Powys has not fallen, and indeed the incidence in children in South Powys may have increased over recent years, although the numbers of children in the surveys were small. The validity of these comparisons depends upon the completeness of ascertainment of diagnosed cases in the two surveys. Thomas's survey involved him personally contacting all hospitals in Wales and 'certain hospitals in England where patients from Wales are admitted for treatment'. He did state that 'one or two fairly large hospitals had not adopted the practice of codifying diseases', and presumably, data from these hospitals were unavailable. Another possible bias which could result in an artificial increase in incidence in childhood is that diagnostic practices and ascertainment of cases improved between studies, although we know of no data to substantiate this suggestion.

In addition to cases in endemic areas, a further 10 patients < 20 years of age with hydatid disease were identified, and three of these lived in the Birmingham area. No obvious source of infection was identified for these patients. Stallbaumer *et al.* (1986) have suggested that the West Midlands cluster could be due to local dogs becoming infected by scavenging carcasses of sheep which have died on farms in the Midlands used to hold Welsh ewes before slaughter. Possibly also, some infected offal may have found its way from slaughterhouses into dog food.

The following recommendations arise out of the study:

(1) Transmission of *E. granulosus* continues to occur in South Powys and Herefordshire, as indicated by the distribution of younger cases, and the hydatid control scheme should be extended to cover the whole of the endemic area including South Herefordshire.

(2) Many people, including doctors, mistakenly believe that human hydatid disease is acquired from sheep. To ensure the continued success of the control programme, a comprehensive health education campaign is needed (Walters, 1986). The major aims of this would be to ensure that dog owners and hunt kennels do not allow dogs to eat raw sheep carcasses, that dogs are treated with

praziquantel at recommended intervals, and that sheep carcasses are safely disposed of as soon as possible (Howells & Taylor, 1980). A supplement to these measures would be to improve personal hygiene of dog owners in order to minimize human ingestion of *E. granulosus* eggs excreted by their dogs.

(3) People buying dogs from Mid-Wales and Herefordshire should check that the dogs have been treated with praziquantel. A certification scheme for dogs bred in endemic areas might be one way to close this route of spread of hydatid disease.

(4) National surveillance of human hydatid disease should be instituted to identify UK-acquired cases and likely sources of infection. Investigation of possible foci of infection should then be carried out in collaboration with the State Veterinary Service.

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