

CASE REPORTS

Hydatid disease with hilar lymphadenopathy

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Summary

Enlargement of bronchopulmonary lymph nodes is unusual in hydatid disease. In the case described the radiographic appearance and the findings at operation suggested pulmonary tuberculosis. Immune mechanisms involved in the response to hydatid disease are discussed.

Hydatid disease with hilar lymphadenopathy

Hydatid cysts develop in the viscera of sheep and man after ingestion of the ova of the tapeworm *Echinococcus granulosus*. The cyst is the larval form of the worm and in sheep-rearing countries including Wales the definitive host is the dog. Cysts may be single or multiple and depending on their position may go unnoticed or cause symptoms due to pressure. If they erode the bronchial tree their contents may be discharged in the sputum. Rupture due to accidental trauma allows leakage of a protein-rich fluid which can cause a severe anaphylactic reaction and if viable daughter cysts are present the parasite may be widely spread.

In endemic areas hydatid disease must always be considered in the differential diagnosis of a round homogeneous or cavity lesion seen on a chest radiograph. A child is now reported who had a pulmonary hydatid cyst associated with unilateral hilar lymphadenopathy, and which radiologically simulated primary tuberculosis. At operation, caseous satellite lesions were encountered which also suggested a tuberculous process, as did the histological findings in these lesions and the pericyst.

Case report

A 9-year-old boy from a semi-rural community had been referred for tonsillectomy because of recurrent upper respiratory infections. He had recently developed a cough and had a single episode

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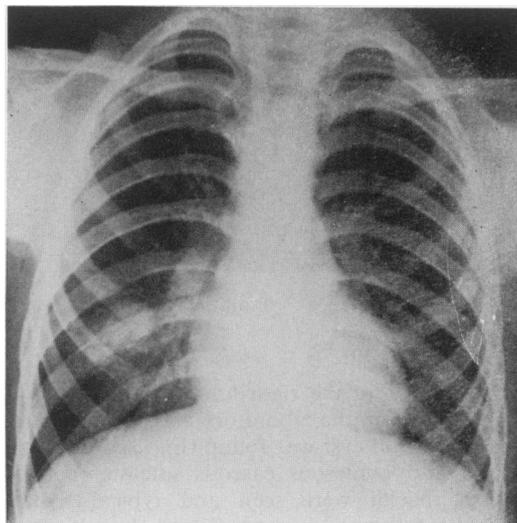


FIG. 1. Circular shadow in the lower lobe of the right lung.

of wheezing and a chest radiograph showed a circular shadow in the lower lobe of the right lung (Fig. 1). There was hilar lymphadenopathy which was confirmed by a tomogram (Fig. 2). He appeared well with no abnormal signs in the chest or abdomen. Investigations included: WCC $9.5 \times 10^9/l$ (eosinophils 3%); ESR 16 mm/hr; Mantoux test 1/1000 negative; precipitins to *Aspergillus fumigatus* and *A. niger* and to *Candida albicans* negative. The Casoni test (Behringwerke) produced an early flare reaction followed by a red area of 4 cm diameter (with induration of approximately 2.5 cm diameter) developing by 24 hr, interpreted as a dual skin response (types I and IV); hydatid CFT (Colindale laboratory) positive at 1/64; immunoglobulins were normal except for an IgE of 1060 i.u./ml (normal <46 i.u./ml). Bronchoscopy showed no abnormality. At thoracotomy there was a mass of hilar glands and when incision of the pleura showed pus-like lesions,

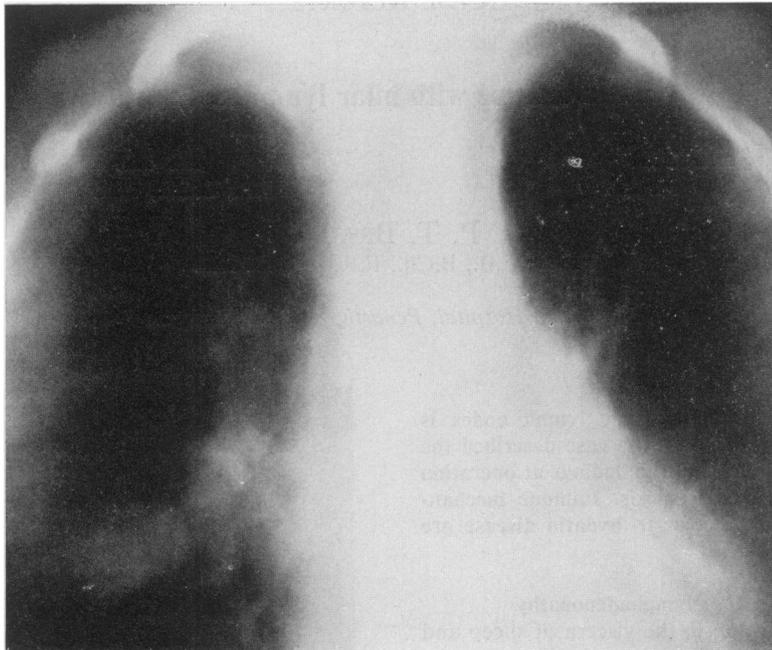


FIG. 2. Film from tomogram showing hilar lymphadenopathy.

the lower lobe of the right lung was removed. On deeper section in the laboratory a 2.5-cm collapsed, infected hydatid cyst was found (Fig. 3) with a thick pericyst and numerous caseous satellite foci. No acid-fast bacilli were seen and typical hydatid hyaline membrane was confirmed histologically. The satellite lesions were bounded by epithelioid and Langhans' giant cells. Hilar nodes showed reactive hyperplasia.

His postoperative course was entirely satisfactory. A technetium scan showed no evidence of hepatic cysts.

Discussion

Although in sheep-farming communities hydatid cysts are usually found in the liver, pulmonary lesions occur in about one third of cases (Howkins, 1966). In the milder disease reported from North America, where the parasite has its life cycle in wolves and deer, the lung is the commonest site and Wilson, Diddams and Rausch (1968) reported that three out of nine patients with ruptured or secondarily infected cysts were thought to have a tuberculous cavity before operation.

The round opacity on the chest radiograph in the present case together with the unilateral hilar enlargement suggested a primary tuberculous complex. Other possibilities included a mycetoma

in a pre-formed cavity, following a tuberculous or pyogenic infection. It seems likely that the hydatid cyst had already ruptured when the initial radiograph was taken and the leak may have caused the episode of wheezing that occurred before the child was first seen. It was remarkable that not only the chest radiograph but the findings at operation suggested tuberculosis and this case illustrates the need to consider the possibility of hydatid disease even when the bronchopulmonary nodes are enlarged.

Factors governing susceptibility to infection with *Echinococcus* are not clearly understood, but the fact that most patients have only a single cyst (Orueta *et al.*, 1974) suggests an active immune response. The positive Casoni test indicates both immediate and delayed type hypersensitivity reactions and it is likely that these mechanisms play some role in the host's defence. Raised concentrations of both serum IgE and specific IgE antibodies are present in hydatid disease (Dessaint *et al.*, 1975) and although parasitic infection can stimulate reaginic response to a variety of previously encountered antigens this non-specific effect is generally short-lived (Orr, Riley and Doe, 1971).

Bronchopulmonary node enlargement is commonly seen in tuberculous and fungal infection of the lung where delayed hypersensitivity mechanisms are known to be important, as well as in sarcoid



FIG. 3. Lobectomy specimen. A 2.5-cm cyst with the collapsed hydatid hyaline membrane on the left. A few small satellite lesions can be identified inferiorly.

where they are defective. However, the reactive hyperplasia seen in the present case was probably due to the secondary bacterial infection often associated with ruptured cysts (Orueta *et al.*, 1974) rather than a specific response to hydatid antigen.

Macrophages are probably involved both as phagocytes and in their co-operative role in the afferent limb of the lymphocyte response. In the alveolar form of hydatid disease they appear to limit seeding and growth of hydatid embryos in rats (Rau and Tanner, 1975).

The presence of complement fixing antibodies which diminish in titre after removal of cysts has some value in assessing the result of surgery (Todorov *et al.*, 1976).

Complicated lesions such as the one described are best treated by pulmonary resection which minimizes the risk of disseminating the parasite or causing anaphylactic shock by spillage of the cyst contents. When cysts are intact it is often possible to remove the ectocyst and its contents successfully either

alone or with part of the fibrous outer capsule. A recent report (Bekhti *et al.*, 1977) suggests that the anthelmintic mebendazole may be effective in destroying cysts which cannot be excized surgically.

The most important factors in the control of hydatid disease are education of the public and prevention of dogs from feeding on raw infected offal. Following these measures the disease has been eradicated from Iceland (Beard, 1973) where the incidence was once the highest in the world.

Acknowledgments

We thank Mr T. Rosser for permission to report this case; Mrs M. Diment for preparing the manuscript; and the Department of Medical Illustration, Llandough Hospital, for the photographs.

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