MEDIASTINAL HYDATID CYSTS

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There are few reports of hydatid cysts of the mediastinum in recent medical literature in English, for in most countries the disease is uncommon. A record of 7 patients with such hydatids in our local experience may therefore be of interest.

Of the 52 mediastinal tumours treated surgically during the last 7 years in the thoracic surgical unit at Groote Schuur Hospital, 7 were due to extrapleural echinococcal cysts—as high a proportion as 13·5%. This number is about 6% of all patients with hydatid disease treated at the hospital during this period, whereas pulmonary cysts formed about 90%.

There are numerous articles on mediastinal tumours with no mention of hydatid cysts amongst the conditions seen. This is true especially for North America but also, for example, for a Scandinavian series of 155 tumours.[1] In the large monograph by Heuer and Andrus published in 1940,[2] 1 case is mentioned which presented as a swelling above the right breast and which was drained externally. Passing mention is also made of 7 cases of hour-glass cysts presenting as spinal tumours, but no clear references are given.

In 1952, in a report from Australia on 26 mediastinal tumours,[3] 1 was a presumptive hydatid; the patient had a hydatid cyst of the liver but refused thoracotomy for a round mediastinal shadow.

Recent literature in Spanish (South American), Russian and Italian contains reports of hydatid cysts of the media-

stinum, pericardium, and heart. In 1954 a good outline of cardiac hydatids[1] with a reportof a case was published in Britain, but in our experience we have not encountered one, though they have been variously reported as from 0·5% to 2% of all hydatid cysts. The 7 cases presented below comprise 5 single hydatids, one case with 2 cysts, and a case with 5 mediastinal cysts and 1 pericardial cyst.

CASE REPORTS

Case 1. *H.F.—White male aged* 27 *years*

A mass was noted at the left apex on mass X-ray examination. He admitted to having had occasional pains in the left chest but this was not a prominent symptom. The mass was ovoid, about 7 cm. across, and medially entered the mediastinum at about the 3rd and 4th ribs. The pre-operative diagnosis was neurofibroma.

At thoracotomy an extrapleural hydatid cyst was found. Unfortunately the cyst ruptured during removal; some formalin was used in the sac and the rest was sucked out. A 6-year follow-up has shown no recurrence.

Case 2. *G.B.—Coloured male aged* 32

He presented a picture almost identical to the previous case, except that the cyst was slightly smaller. It was removed without rupture.

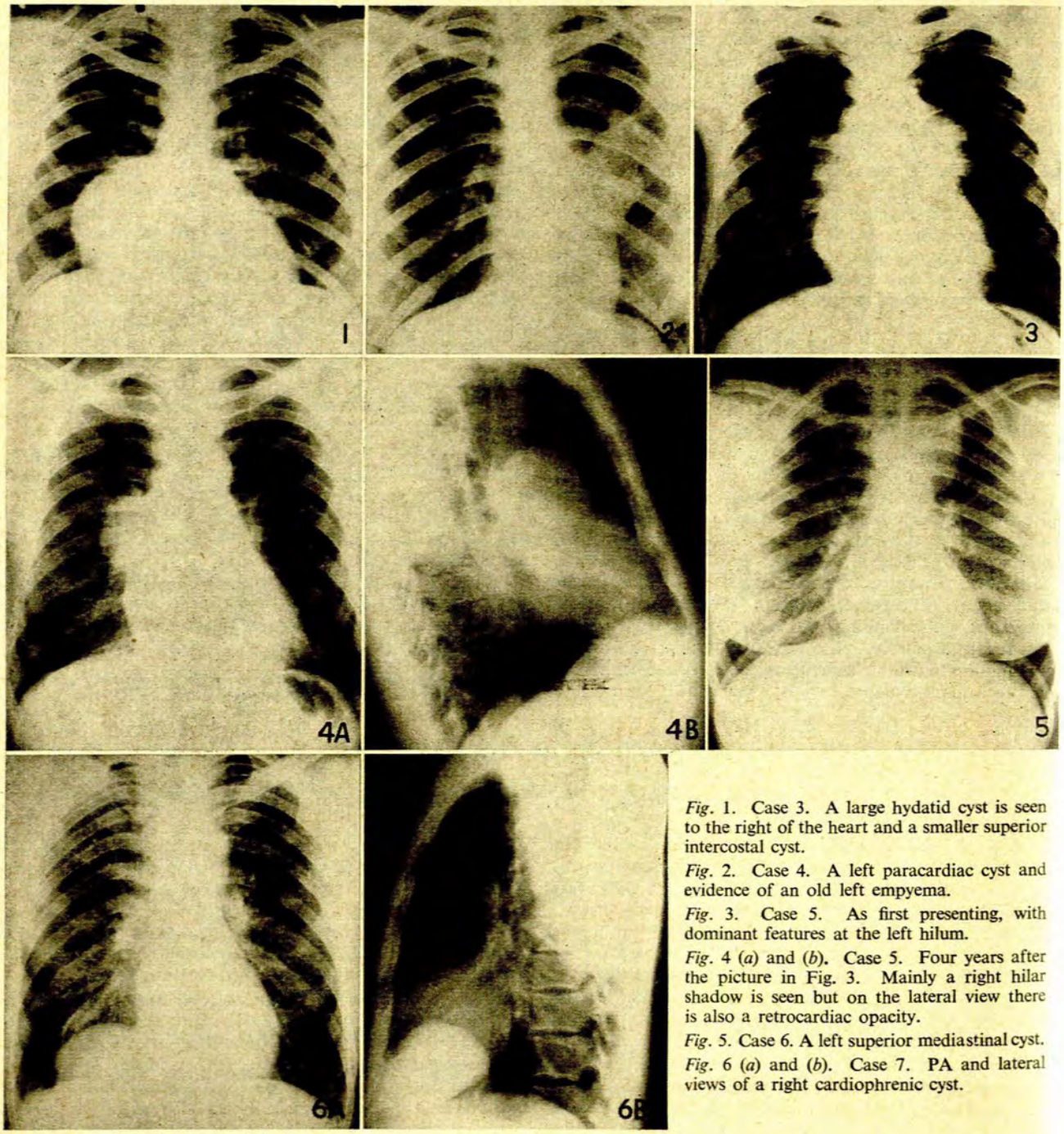

Fig. 1. Case 3. A large hydatid cyst is seen to the right of the heart and a smaller superior intercostal cyst.

Fig. 2. Case 4. A left paracardiac cyst and evidence of an old left empyema.

Fig. 3. Case 5. As first presenting, with dominant features at the left hilum.

Fig. 4 (*a*) and (*b*). Case 5. Four years after the picture in Fig. 3. Mainly a right hilar shadow is seen but on the lateral view there is also a retrocardiac opacity.

Fig. 5. Case 6. A left superior mediastinal cyst.

Fig. 6 (*a*) and (*b*). Case 7. PA and lateral views of a right cardiophrenic cyst.

Case 3. *S.V.R.—White female aged* 28

Largely asymptomatic, she admitted to a feeling of heaviness in the chest, and on radiological examination 2 well-defined round opacities were visible in the right chest (Fig. 1). The larger was about 7 cm. in diameter and lay against the right border of the heart; the other was about 3 cm. across and projected from the apex of the thoracic space. Hydatid cysts were suspected, and this diagnosis was confirmed at thoracotomy. The larger cyst was lying under the mediastinal pleura adjacent to the pericardium and the smaller was situated extrapleurally in the second intercostal space. Both were removed without difficulty.

Case 4. *P.F.—White female aged* 22

When about 3 years old this patient had had an empyema of the left chest drained, and when we obtained the doctor's report it appeared that this had been caused by a hydatid, for 'grape skins' had drained with the pus. This empyema healed satisfactorily and apparently left her with no disability.

She trained as a nurse and, though a small mediastinal swelling was then noted on X-ray, the radiologist associated this with scarring from the old empyema and she was not referred to a thoracic surgeon until after finishing her training.

When seen, she was asymptomatic, but a cystic swelling was seen projecting from the left hilar region on the P.A. film, ovoid and about 6 cm. across (Fig. 2). The lateral view showed this to lie in the anterior mediastinum, and a dermoid was suspected. At thoracotomy, there was some scarring and adhesion from the old empyema, and a live hydatid was removed from the anterior mediastinum without rupture.

Case 5. *J.M.—Bantu male aged* 29

Mass X-ray revealed an asymptomatic lesion at the left hilum. It was lobulated but well-defined and lay just anterior to the pulmonary structures (Fig. 3). There was possibly a small increase in the size of the right hilum also and disease of the lymphatic glands was suspected. There were no other glands or findings elsewhere to aid in the diagnosis, and so diagnostic thoracotomy was performed. Two hydatid cysts were found in the mediastinum just anterior to the hilum, and these were successfully removed.

Further radiological check (Fig. 4, *a* and *b*) showed the lesion at the right hilum to increase in size and about 4 years later a second thoracotomy on the right was performed. During this period the cyst had grown to about 6 cm. in diameter; presumably it had been perhaps 2 cm. when first seen. At thoracotomy the cyst visible on the X-ray film was found to be a live hydatid displacing the phrenic nerve laterally and coming out from between the superior vena cava and the right pulmonary artery; it was evacuated *in toto* from within the ectocyst, which was adherent to the vessels. The pericardium was adherent and slightly thickened; after it was opened an inspissated hydatid cyst was found in relation to the superior pulmonary vein within the pericardial cavity, which was obliterated. Finally 2 more inspissated cysts were found just outside the pericardium in relation to the inferior pulmonary vein. These inspissated cysts did not obviously appear to be infected and the reason for their death can only be speculative, presumably associated with the heartbeat. Convalescence was normal.

Case 6. *L.V.R.—White female, aged* 24

Again an asymptomatic cyst lying on the left side of the superior mediastinum was found on routine X-ray (see Fig. 5). In contrast to cases 1 and 2, the longitudinal diameter was vertical and there was some lobulation. There was no widening of an intercostal space. The hydatid cyst was lying extrapleurally at the medial end of the third intercostal space. Unfortunately this cyst was also ruptured during removal.

Case 7. *B.C.—Coloured male, aged* 48

Routine mass radiographic examination showed this patient to be harbouring a symptomless cyst lying in the right anterior cardiophrenic region (see Fig. 6, *a* and *b*). At thoracotomy, the cyst was found to be extrapleurally situated and to arise from the diaphragm without any hepatic connection. The cyst was readily removed *in toto*.

DISCUSSION

The route of infection in these cases is somewhat difficult to explain except on a fortuitous basis. The usually accepted route of infection in hydatid disease is from the bowel to the liver as the first filter, where perhaps 50% of ova settle, particularly the larger ones; thence to the lungs, where some 25% are filtered out; and finally to the heart by the coronary vessels and to the body in general. An alternative entrance might be by direct inhalation of ova in dust to the lungs. By either route, it would be reasonable to expect pulmonary hydatids in association with mediastinal ones and D'Abreu[5] in his book makes a brief note that these cysts are 'almost invariably secondary to hydatid disease of the lung'. In this series of 7 cases, however, there was only one in which a presumptive diagnosis of pulmonary hydatid cyst could be made in retrospect (case 4), which tends to rule out metastatic venous secondary echinococcosis. In the case with 6 cysts there was no evidence of pulmonary disease. Thus the possibility of lymphatic spread from the bowel *via* the thoracic duct must be borne in mind. The mediastinal cysts may be either anterior or posterior, so that lymphatic spread in all cases is unlikely; but 3 of the patients in this series had single cysts high on the left and posteriorly. Possibly cardiac and aortic pulsation may prevent development lower along the thoracic duct; though, in case 5, 2 of the cysts in relation to the pulmonary ligament would be close to the duct and might have developed from it. It would be difficult to postulate lymphatic spread for the other cases and these must presumably be haematogenous, settling fortuitously in the mediastinum. However, the question has been raised in the past whether the preponderance of hepatic and pulmonary cysts does not indicate some organ affinity rather than mere filtration from the blood. No answer to this possibility has as yet been offered.

With multiple cysts one is tempted to consider that there was an original single cyst which ruptured in a mobile mediastinum, owing either to cardiac pulsation or to external trauma. This is said usually to be the case with pericardial cysts, which develop as a result of the rupture of a cardiac cyst. In case 5, however, there seemed to be only 1 pericardial cyst and there was no evidence of a cardiac one. The pericardial cavity was completely obliterated, as apparently is often the case; a certain degree of constriction of the pericardium has been reported, attributable to the irritant effects of the hydatids, but it was not evident here. Solitary pericardial cysts are uncommon, but they may be associated with pain or symptoms caused by pericardial effusion.

Two of these cysts were inadvertently ruptured during removal, but no recurrence has been noted, in one case over a period of 6 years. This accords with our experience with pulmonary cysts, where we have seen no recurrences although cysts have ruptured during removal. The reason seems to lie in the fact that the 'thin' cysts, which are so liable to rupture with the slightest manipulation, are in fact undisturbed acephalocysts lacking the stimuli to the formation of brood capsules or scolices. According to Dew,[6] who stressed the persistence of the parasitic elements, any of these may, if shed into the tissues under aseptic conditions, implant themselves and give rise to secondary cysts. There seems little doubt that fragments of the original germinal membrane may also lay down protective laminated membrane around islets of nuclear material and may give rise to cysts. These secondary cysts, however, are of very slow growth and may take from 5 to 12 years to be obviously manifested, thus lulling the surgeon into optimistic misinterpretation. It may be that that is the case in the series reported here.

The rate of growth is illustrated to some extent in these

cases. All presented in early adult life, which suggests that infection took place at an early age. Case 4 seems to have already had a cyst in the lung at the age of 3, although the mediastinal one was only of medium size 2 decades later. In case 5, the radiological growth was in the region of 1 cm. per year during observation. One cannot but feel that the rate of growth varies from cyst to cyst and even from time to time in the same cyst, perhaps according to the blood supply.

SUMMARY

Seven cases of hydatid disease of the mediastinum are reported, seen in recent years at the thoracic surgical unit of Groote Schuur Hospital. One intrapericardial cyst is included.

The possible route of infection and rate of growth are discussed and some of the literature referred to.

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